



Parental stress and child adherence to treatment plans: A systematic review

**A distance-based intervention supporting neuropsychological
recommendations for children with a neurodisability**

Submitted by Dr Jessica Watts, to the University of Exeter
as a thesis for the degree of Doctor of Clinical Psychology, December 2019

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I certify that all material in this thesis which is not my own work has been identified and that no material has previously been submitted and approved for the award of a degree by this or any other University.

Signature:

A handwritten signature in purple ink, appearing to read "J. Watts", written over a horizontal line.

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SCHOOL OF PSYCHOLOGY

DOCTORATE IN CLINICAL PSYCHOLOGY

LITERATURE REVIEW

Parental stress and child adherence to treatment plans: A systematic review

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Abstract

Background: Adherence to treatment plans for children with chronic disorders is around 50%. Barriers to good adherence include parental factors such as parental stress, with parents of children with a chronic condition found to have higher levels of parental stress than parents of children without such a condition. However, no review has assessed the relationship between parental stress and adherence irrespective of child diagnosis.

Method: PRISMA-P guidelines were followed to ensure transparency. CINAHL, Medline, EMBASE and PsycINFO databases were searched using relevant terms from creation to July 2019. 1067 articles were identified and screened for eligibility, resulting in 14 studies being included.

Results: Overall a negative relationship between parental stress and adherence was found, such that increased parental stress related to poorer adherence. Exceptions to this are discussed. Papers utilised varied measures for parental stress and adherence and assessed a number of different childhood disorders. Parental stress was found to be a multifaceted concept including aspects such as time pressures, emotional strain and financial difficulties.

Conclusion: Parental stress is a possible target for interventions aiming to improve paediatric adherence. However, given the multifaceted nature of parental stress interventions may benefit from targeting particular aspects (e.g. alleviating financial pressures). Further, clinics could work towards routinely screening for parental stress and developing a clinical cut-off indicating high stress among parents whose child has a chronic illness. Future

research directions include further consideration of additional factors identified within this review as also related to adherence (e.g. health beliefs, family conflict).

Keywords: parental stress, paediatric chronic health disorder, adherence

Introduction

Improved understanding and disease management has led to improved survival for chronic illnesses (Halfon & Newacheck, 2010; Mookink, van der Lee, Grootenhuys, Offringa, & Heymans, 2008). However, individuals and their families experience burden related to the disorder (e.g. managing news of the diagnosis, health appointments and care needs as well as maintaining family life). Therefore, parenting and caring for a child with a chronic health condition can bring parents stress. Research has understood the presence of this stress for nearly 30 years (Abidin, 1990); however, how this stress affects families and parental management of the chronic illness is poorly understood.

The number of families affected by paediatric chronic illness underscores the importance of understanding the role of parental stress. Whilst statistics are collected on the prevalence of adult chronic illness, the same focus has not been placed on paediatric conditions within the UK (NHS Digital, 2016). Instead, prevalence rates are determined for specific illnesses, with recent estimates for three of the most recognised paediatric conditions in the UK being: 1 in 11 children for asthma (Asthma UK, 2019), 1 in 700-1000 for Type 1 Diabetes (Diabetes UK, 2010) and 1.37 per 10,000 for Cystic Fibrosis (Farrell, 2008). Therefore, although it is difficult to determine an overall estimate of the prevalence for paediatric chronic illnesses within the UK, it is clear the conditions are widespread. Further, worldwide the prevalence of paediatric chronic illnesses is on the rise (Asher et al., 2006).

Healthcare for these children and families is tasked with developing treatments that can enable the child to live as 'typical' a life as possible.

Children are prescribed multifaceted treatment plans, which can include medications but also completing physiotherapy (e.g. for Cystic Fibrosis, CF), routine blood glucose checks (e.g. for diabetes), frequent hospital visits, lifestyle recommendations (e.g. changes in diet) and for children where their cognition may be impacted, educational or learning plans. As the disorders are chronic, these plans are also long-term. Much of this treatment can be completed at home and as such the onus on families to travel is reduced. However, the impact of having treatment that can be completed at home means care is often managed by parents or caregivers. Good adherence therefore involves the parents, child and the professionals who provide the plan (De Civita & Dobkin, 2004).

Poor adherence is associated with poor disease control, reduced quality of life (McGrady & Hommel, 2016) and in extreme circumstances, mortality (Suissa, Ernst, Benayoun, Baltzan, & Cai, 2000). However, adherence across chronic disorders is typically around 50% and reduces in developing countries World Health Organisation (WHO, 2003) and during adolescence as children take more responsibility for their self-care (Taddeo, Egedy, & Frappier, 2008). Not only is there a direct cost to the child of reduced health, but there is also an economic impact with costs of providing emergency care or scheduling additional medical appointments being estimated to each >\$300billion in America (McGrady & Hommel, 2016). Further, the WHO, estimate that chronic diseases will account for 65% of global disease burden by 2020 (WHO, 2003). Understanding adherence and

in turn tailoring interventions to improve adherence among children is therefore crucial. Rather than needing to develop more targeted and specific medical treatments, the WHO cite research that suggests improving adherence may have greater benefit to reducing the burden of long-term conditions (Haynes, McDonald, & Montague, 2002; WHO, 2003).

Research has identified multiple parental factors that affect adherence across chronic paediatric disorders, such as parental mental health or distress (Cline, Schwartz, Axelrad, & Anderson, 2011; Horsch & McManus, 2014; Sheehan et al., 2012; Whittemore, Jaser, Chao, Jang, & Grey, 2012), parental resources (Dunst, Leet, & Trivette, 1988; Happ, Hoffman, DiVirgilio, Higgins, & Orenstein, 2013), family structure or density (Caccavale, Weaver, Chen, Streisand, & Holmes, 2015; Chan et al., 2016; Dashiff, Bartolucci, Wallander, & Abdullatif, 2005), family functioning (Drotar & Bonner, 2009; Kokkonen, Taanila, & Kokkonen, 1997), parenting style (Radcliff, Weaver, Chen, Streisand, & Holmes, 2018; Robinson, Weaver, Chen, Streisand, & Holmes, 2016), and parental stress (Robinson et al., 2016). The number of these factors indicate researchers are aware of the need to better understand adherence, but also highlight the complexity. Given the poor levels of adherence among paediatric chronic illnesses and the impact this can have, the importance of better understanding the relationship between parental factors and adherence is clear. Parental stress has been repeatedly identified in research among childhood chronic illnesses (Cousino & Hazen, 2013; Robinson et al., 2016; Streisand, Braniecki, Tercyak, & Kazak, 2001) and is therefore potentially a key issue.

The nature of how factors impact adherence is complex though, with varied definitions of adherence (e.g. following a prescribed treatment or a health outcome such as glycaemic control) and research often investigating one factor within one disorder. Although such focus is understandable due to the nature of research grants and enhancing interpretation of results through a homogenous sample, the knock-on effect is difficulty in unpicking the overall impact of one factor (e.g. parental stress). Researchers and clinicians would therefore benefit from drawing together understanding across multiple disorders and utilising a framework or model to design and interpret results.

Models relevant here include those relating to paediatric self-management and adherence that also reference the importance of family and societal systems (e.g. the Circumplex Model of Marital and Family Systems, Olson, 2000; and the Pediatric Psychosocial Preventative Health Model [PPPHM], Kazak, 2006). The strengths of these models include clearly identifying, conceptualising and measuring family factors (e.g. communication, flexibility and cohesion within Olson's model) and being able to include a spectrum of families, ranging from those where children do not have health needs to those that do (the PPPHM model). However, a third model (Modi et al., 2012) is particularly relevant here not only because it covers the above aspects but also because it clearly defines key terms, aiding use and interpretation. The pediatric self-management framework by Modi et al., (2012) utilises the ecological systems theory, acknowledging behaviours occur within four domains: child, family, community and the health care system. Self-management behaviours are identified as influenced by various processes within these domains, that are either modifiable or not (e.g.

psychological or cognitive factors and health care provider communication). This model provides a clear framework and rationale for research related to paediatric chronic health conditions and how to target interventions at processes that are modifiable.

Modi and colleagues' framework (2012) enables understanding to develop regarding transdiagnostic factors, such as parental stress, on adherence. This approach has been supported by recent reviews, which attempted to draw conclusions across disorders (Cousino & Hazen, 2013; Psihogios et al., 2019). Psihogios et al. (2019) found that various aspects of family functioning were related to reduced adherence (e.g. greater family conflict, lower cohesion, reduced family flexibility, more negative communication and poorer problem-solving). Although this meta-analysis considered multiple conditions (e.g. asthma, CF, epilepsy, sickle cell disease) and conceptualised family functioning along a variety of measures, an indication of the level of parental stress was not investigated. Cousino & Hazen (2013) reviewed studies that measured parental stress among caregivers of children with a chronic condition. Their findings showed that parents of a child with a chronic illness experience greater levels of parental stress than parents of healthy children. Further, parental stress was negatively associated with positive parental cognitive appraisals about their child's illness and parental self-efficacy of disease management. However, parental stress was positively related to parental responsibility for their child's treatment (e.g. parents reported more stress when they felt more responsible for managing their child's care). Although the review did not assess the impact of increased parental stress on adherence, the authors did find that parental

stress was related to poorer psychological adjustment (Hilliard, Monaghan, Cogen, & Streisand, 2011; Kazak & Barakat, 1997) and may contribute to child health outcomes (Barakat et al., 2007). Therefore, interventions to reduce parental stress were identified as a route to improve child outcomes, via adherence. In relation to the pediatric self-management framework (Modi et al., 2012), parental stress can therefore be conceptualised as a modifiable factor acting at the family level via a process of stress management (or non-management). However, in order to design such interventions, the nature of the relationship between parental stress and adherence needs investigation. Although individual studies have assessed this relationship, to date no review has drawn together this research across paediatric chronic disorders.

This systematic review therefore aimed to collate research assessing the relationship between parental stress among parents of children with a chronic illness and their child's adherence levels.

Method

Preferred Reporting Items for Systematic review and Meta-Analysis Protocols (PRISMA-P; Moher et al., 2015) guidelines were used to ensure a transparent and unbiased methodology. Papers which assessed parental stress specifically in relation to a child's chronic illness and its impact on the family's ability to complete recommendations or treatment prescribed by health care professionals were reviewed.

Eligibility Criteria

As recommended in PRISMA-P (Moher et al., 2015) the Population, Intervention, Comparison, Outcome and Study type (PICOS) framework was used to establish inclusion and exclusion criteria for this review (see Table 1).

This study defined the key concepts of parental stress and adherence as:

- Parental stress: stress experienced by parents related to their child’s chronic health condition, or general stress experienced by parents, but which was shown within the paper to link to their child’s adherence.
- Adherence: the extent to which a person’s behaviour coincides with medical or health advice (as defined in Modi et al., 2012).

Table 1
PICOS framework for review inclusion and exclusion criteria

	Inclusion criteria	Exclusion criteria
Population	Assessed parents of children with a chronic health disorder. Studies did not have to specifically collect data from the children. A chronic disorder was identified through the need for years of treatment (e.g. cancer, epilepsy, asthma).	Studies where parents were not included or where a child’s disorder was not chronic. If a chronic disorder was assessed but in relation to acute emergency medical care, these studies were also excluded.

	Inclusion criteria	Exclusion criteria
Intervention	<p>Studies had to measure parental stress as related to their child's disorder (e.g. either a specific quantitative measure of stress or a qualitative interview where burden or stressors were attributable to the child's disorder). N. B. Questionnaires not specifically designed to measure parental stress in relation to a child's chronic disorder were eligible when the study's aim was to better understand parental stress within the context of a child's chronic disorder. Further, studies were included if descriptive statistics were provided on reasons for non-adherence, only when parental stress was noted.</p>	<p>The following factors could be reported on in the papers, but parental stress could not only be related to:</p> <ul style="list-style-type: none"> • Mental health difficulties for parent or child • Conflict in the family or parent-child dyad • Support in family • Family structure • Family communication style.

	Inclusion criteria	Exclusion criteria
Comparison	Not applicable.	Not applicable.
Outcome	<p>Compliance or adherence to recommendations, treatment or medication.</p> <p>This could include physical therapy, establishing a home routine, implementing educational plans, monitoring child's glucose levels or adhering to a diet.</p> <p>Qualitative studies do not need an independent measure of adherence.</p>	<ul style="list-style-type: none"> • Appointment attendance • Actual medical outcome (e.g. metabolic control) as these could be influenced by factors other than adherence to recommendations.
Study type	<p>If multiple articles were produced from one study the article included in this review was determined based on the inclusion of findings specific to this review question.</p> <p>Peer reviewed articles.</p> <p>Quantitative or qualitative research.</p>	<p>Commentaries.</p> <p>Editorials.</p> <p>Non-English articles.</p> <p>Books.</p> <p>Reviews.</p>

Key: PICOS = Population, Intervention, Comparison, Outcome and Study type

Search Strategy and Sources

Search terms and strategy were developed in collaboration with researchers and librarians, through identifying relevant reviews (Cousino & Hazan, 2013; Drotar & Bonner, 2009; Whittemore et al., 2012) and the use of pilot searches. Although not used in all the aforementioned reviews, truncated terms were piloted when refining the search strategy (e.g. parent* stress). However, they did not increase the identification of mapped terms or relevant papers and were not used in this review. The following databases were searched from creation to July 2019: OVID Medline (R), PsycINFO, EMBASE and CINAHL.

MEDical Subject Headings (MESH) terms were mapped onto relevant terms. Below is an overview of the MESH terms and strategy for all databases. Notably, each database indexes articles independently, leading to variable mapped terms. A full list of mapped terms for each database can be found in Appendix A. All searches had the following restrictions: articles, human and English language. To ensure all relevant papers were included, reference lists were screened. Grey literature was excluded from this review due to time constraint.

Of note, in EMBASE 'adherence' did not yield appropriate mapped terms and restricted the number of articles identified and therefore 'non-adherence' was used as an alternative. Finally, in PsycINFO 'adherence' and 'non-adherence' were used to maximise search specificity.

Search terms used:

- (Child) OR (Pediatric) OR (Adolescent) AND
- Parental stress AND
- Chronic Disease AND
- Adherence (or/and non-adherence) AND
- Health Behaviour

Health behaviour was included in the search to capture information related to adherence of all recommendations (i.e. not those solely related to medical treatment).

Data Collection

References were collated into Endnote for screening and the removal of duplicates. The full screening process is laid out in Figure 1. PICOS inclusion criteria were used to screen article titles and abstracts. All remaining articles (n = 153) were read in full, resulting in 14 articles being included in the final review. Six of the 153 articles were randomly selected and rated blind by another researcher using the PICOS criteria. Disagreements were discussed and criteria clarified, leading to 100% inter-rater reliability for inclusion and exclusion criteria.

Study Quality and bias

Tools recommended within the National Institute for health and Care Excellence (NICE) guidelines (NICE, 2019) were trialled when choosing appropriate quality appraisal tools for this review.

The quality of the three qualitative studies included was judged using the Critical Appraisal Skills Programme (CASP) qualitative checklist (CASP, 2018), see Appendix B. This tool contains 10 items, which cover the validity, results and applicability of the findings. This tool was considered to be

appropriate and feasible for this review as it included appraisal of methodology, design, whether the researcher assessed their biases, ethical considerations and appropriateness of analysis.

The 11 quantitative studies were cross-sectional surveys. Despite a recent call for clarity on how best to critically appraise such studies (Protogerou & Hagger, 2018) the current recommendation from NICE (NICE, 2019) is the 12 item Centre for Evidence Based Management (CEBM) tool, as adapted from a critical appraisal text book (Crombie, 1996) see Appendix C. This tool was found to be most applicable to the 11 quantitative papers in this review as it covered items related to design, sampling, power, response rates, significance and interpretation of results. Each of these questions could be answered for each paper and highlighted areas of strengths and weakness across the included papers.

Although neither scale refers to a scoring strategy, for ease of comparison in this review the two tools scored 'yes' as '2', 'can't tell' as '1' and 'no' as '0'. Two items on the CEBM are reverse scored (items 4 and 11) as 'no' responses indicate a higher quality aspect of the study. It should be noted the total possible scores vary with CASP maximum being 20 and CEBM being 24. For the purpose of this review studies with CEBM or CASP scores of ≥ 17 were considered strong, 14-16 moderate or < 14 weak.

Appendix D contains CEBM and CASP scores. One qualitative and two quantitative articles were second rated, achieving a Cohen's kappa of 0.521, $p < 0.0005$ suggesting a moderate agreement. Differences in quality ratings typically related to how applicable results were to the current review. The second rater was not involved in the review design and was not an expert in

this field. On discussion, in all instances, it was agreed the review author's scores should be maintained. No study was excluded due to quality, but quality scores informed the discussion of findings.

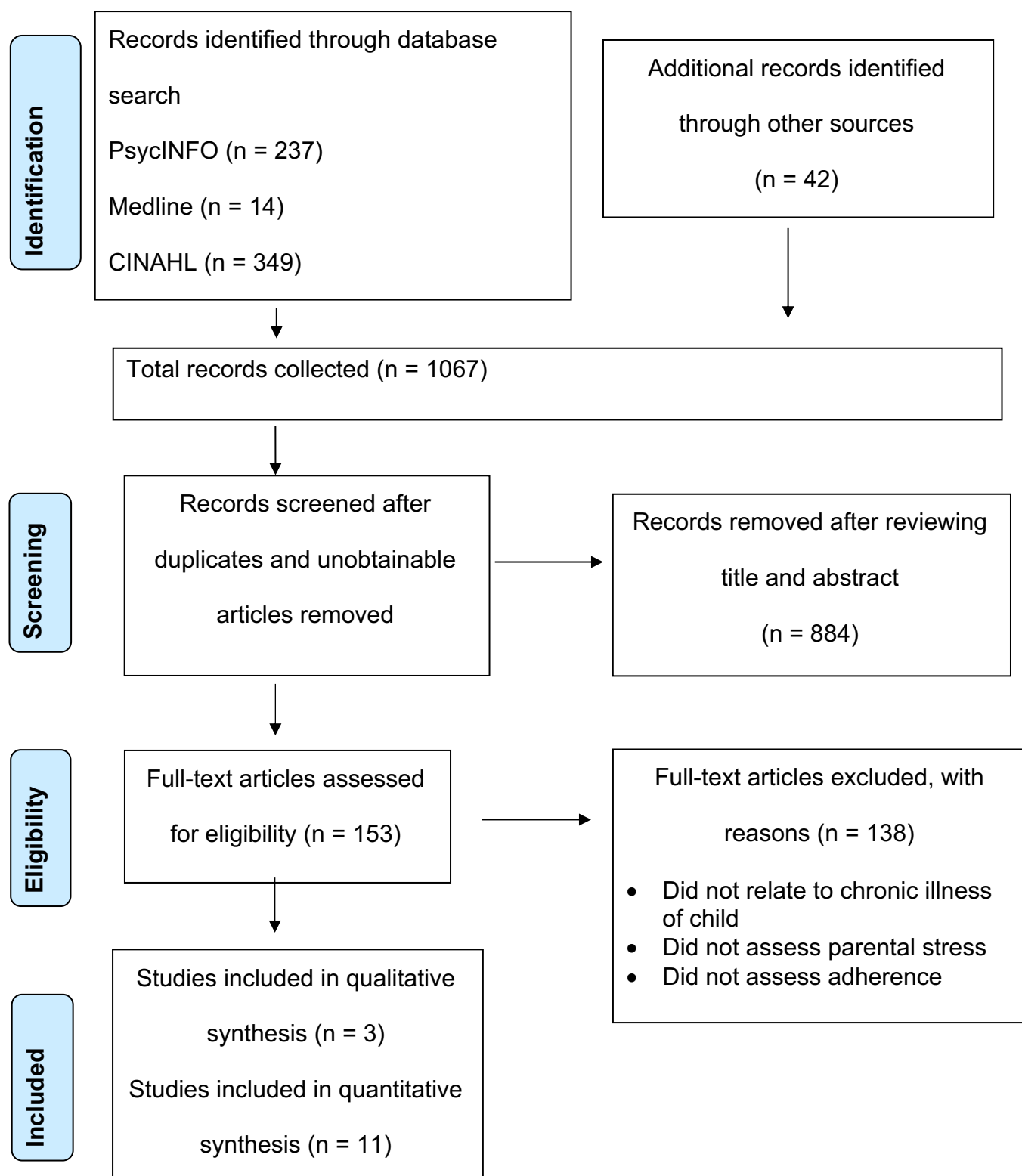
Results

The search

The search and screening process can be seen in Figure 1. Database and reference searches yielded 1067 articles, of which 21 were duplicates. 1038 unique articles were assessed for possible inclusion in this review. Screening of abstracts and titles resulted in 884 articles being removed. Main exclusion reasons were articles not assessing adherence ($n = 283$), not relating to a paediatric disorder ($n = 217$), the disorder not being chronic ($n = 112$) or an absence of parental stress measurement ($n = 108$). 153 articles were read in full to assess eligibility. At this stage the main exclusion reasons were not including parental stress ($n = 97$), not considering adherence ($n = 19$) and the disorder not being chronic ($n = 16$). Fourteen articles were included in this review (Auslander, Thompson, Dreitzer, & Santiago, 1997; Bourdeau, Mullins, Carpentier, Colletti, & Wolfe-Christensen, 2007; Britton, 1999; Burgess, Sly, Morawska, & Devadason, 2008; Celano, Klinnert, Holsey, & McQuaid, 2011; Chaney & Peterson, 1989; Chisholm et al., 2007; DeMore, Adams, Wilson, & Hogan, 2005; Eddy et al., 1998; Klok, Lubbers, Kaptein, & Brand, 2014; McElroy, Konde-Lule, Neema, & Gitta, 2007; Njuguna et al., 2015; Robinson et al., 2016; Rone-Adams, Stern, & Walker, 2004), the details of which can be found in Table 2. From this point onwards, the Table 2 reference number is used to refer to articles.

All articles that considered the impact of parental stress on adherence to treatment for a child with a chronic disorder were included. This relationship was measured in a variety of ways, leading to a heterogeneous sample of studies for this review. Pertinent study characteristics and dominant differences between studies will be considered, followed by the review findings.

Figure 1

PRISMA-P flowchart detailing systematic review search and inclusion process

Study characteristics

Articles were published between 1989 and 2016, indicating this has long been an area interest. However, the inclusion of only 14 articles, suggests a dearth of research specifically addressing the question of whether parental stress impacts adherence for children with a chronic disorder. Of the 14 included studies, 11 were quantitative studies with a cross sectional survey design (1, 2, 4, 5, 6, 7, 8, 9, 12, 13, 14). The remaining three used qualitative assessments (interviews) to determine the link between parental stress and adherence (3, 10, 11). Notably, two of these studies were mixed methods, using quantitative measures of adherence (3, 10).

Studies were conducted across the globe, predominantly in the USA (1, 2, 5, 6, 8, 9, 13, 14) with remaining studies being distributed between the UK (3, 7), Australia, (4), Holland (10), Uganda (11) and Kenya (12). Sample sizes varied between relatively small (e.g. between 20 and 30 participants [6, 10] to samples of over 100 families [1, 2, 13]). Eleven studies included parent – child dyads (1, 2, 3, 4, 5, 6, 7, 8, 9, 13, 14). The remaining three studies included information about the child, but only collected responses from the parents of a child with a chronic disorder (11, 12, 13). Thirteen studies examined one disorder each: Type 1 Diabetes (T1D; 1, 14), juvenile arthritis (3, 6), Cystic Fibrosis (CF; 9), asthma (4, 5, 8, 10), Clubfoot (11), cancer (12) and neuromuscular disorders (14). However, the final study assessed asthma, CF and T1D (2).

Across all 14 studies, 1172 parents were sampled, with the predominant respondents being mothers. The age of parents was reported in

three studies (1, 2, 5). In the studies that recruited parent-child dyads (1, 2, 3, 4, 6, 7, 8, 9, 10, 13, 14), 997 children were included, with 552 being female.

Two studies (11 and 12) were conducted in the developing world and discuss findings relevant to those specific cultures (e.g. the impact of hierarchy and bewitching practices). However, the parental stress findings did not represent culturally specific factors and therefore they will be considered in conjunction with all other studies.

Of note, three studies (5, 10, 13) reported being part of a larger study. The included studies presented findings specific to the current review aim. No two studies in this review involved participants from the same cohort.

Study Quality

The CEBM scores for quantitative studies indicated that four studies received a quality score of 'strong' (1, 5, 7, 13), five scored 'moderate' (2, 6, 8, 9, 13) and two scored 'weak' (4, 14; Table 5 in Appendix D). Only one study reported power calculations in determining their sample size (4) and therefore whilst some studies had samples of over 70 (1, 2, 13, 14), their ability to appropriately complete their statistics is unclear. All studies provided clear aims and utilised appropriate designs for their objectives. The main areas of weakness for studies was the absence of Confidence Intervals (CIs; 1, 2, 4, 5, 6, 7, 8, 9, 12, 13, 14), uncertainty about how representative the sample was (1, 2, 4, 6, 7, 8, 14) and the lack of confound considerations in analysis (1, 2, 4, 6, 8, 9, 13, 14). In addition to these issues, the two studies with 'weaker' CEBM scores in particular failed to articulate the recruitment process (4), and had low or unclear response rates (4, 14). They recruited 117 parent-child

dyads with asthma (4) and neuromuscular disorders (14). These studies will be included in the review, but their findings will be given less weight than the other studies. The strongest studies were those with large samples, clear recruitment processes and thorough analyses (1, 7, 13).

The quality range of qualitative studies was smaller, with CASP scores of 17 (3), 18 (10) and 19 (11) out of a possible 20, therefore all studies scored within the 'strong' range (Table 6 in Appendix E). Studies reported clear aims, methods and generally considered both ethical factors (10, 11), and the impact of the researcher well (3, 10, 11). However, the thoroughness of analysis was an area of issue for most studies (3, 10, 11), as theoretical underpinnings were not clearly elaborated.

Overall, the quality of 12 of the included studies were scored as 'moderate' or 'strong' (1, 2, 3, 5, 6, 7, 8, 9, 10, 11, 12, 13). The results from these will be weighted more heavily when determining generalisability of review findings.

Table 2

Summary and main findings of articles included in systematic review. Ordered alphabetically by first author.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
1. Auslander et al., 1997 USA	Cross sectional survey Examine factors related to mother's satisfaction with care and if this predicts adherence and medical outcomes.	158 children with T1D & mothers, recruited from university clinics. Children: 50% female, mean age = 12.6; SD = 3.5	FILE (McCubbin & Patterson, 1987) Also measured community stress using portion of SIS (Dressler, 1991) and family resources using FIRM	Adherence & IDDM Questionnaire- R (Hanson, Henggeler, & Burghen, 1987). Measured adherence to diet, hypoglycaemia treatment &	Pearson correlation & multiple regression.	FILE score in normative range. Mean adherence = 24.1 (parent, total) and 22.6 (child, total). Parental stress & community stressors (e.g. racism) related to mother's satisfaction with	CEBM = 17/24 (strong). Strengths: <ul style="list-style-type: none"> • Two recruitment sites • Large sample size • Strong analysis • Inclusion of confounders (e.g. perception of racism) • Adherence operationalised in

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		Mothers: mean age = 39.3; SD = 7.2 Diabetes present for ≥1yr & child under 18 years old.	(McCubbin & Comeau, 1987).	glucose & urine testing.		care. Satisfaction affects adherence, which impacts metabolic control. No direct link between parental satisfaction & metabolic control.	multiple ways (e.g. diet, glucose and urine testing) • Separate adherence & metabolic control. Limitations: • Cross-sectional design • Sample size not based on power calculation • Unknown response rate to study invitation

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
							<ul style="list-style-type: none"> • Potentially biased sample with mainly white & two parent families. • Possible link between parental stress & adherence only assessed via mother's satisfaction with care.
2. Bourdeau et al., 2007	Assess relationship between 1) parenting	200 parent- child dyads Children had either	PSI-SF (Abidin, 1990).	SCI (La Greca, 1992, 2004) developed for diabetes and	Correlation and hierarchical multiple	Parenting stress associated with lower parent ratings of child	CEBM = 15/24 (moderate). Strengths:

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
USA	stress, 2) parental overprotectio n and 3) perceived child vulnerability to child self- care.	T1D (n=124), asthma (n=48) or CF (n=29) Mean age = 12.3, SD = 2.8.. Mother's mean age = 39.6, SD = 5.2. Father's mean age =	Also measured perceived child vulnerability with CVS (Forsyth, Horwitz, Leventhal, Burger, & Leaf, 1996) parental overprotection with PPS (Thomasgard, Shonkoff, Metz, & Edelbrock, 1995)	adapted for asthma and CF for this study. Rated by all parents and children over 8 years old.	regression analysis.	self-care ($\beta = -.19$, $p = .01$) and near significant association with child ratings of self-care ($\beta = -.14$, $p = .07$) for all disorders combined. Non-significant associations in analysis for individual disorders.	<ul style="list-style-type: none"> • Large overall sample size • Thorough analysis • Inclusion of multiple disorders • Inclusion of mothers and fathers. Limitations: <ul style="list-style-type: none"> • Cross-sectional design • Sample size not based on power calculation

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		41.9, SD = 6.5. Recruited from three clinics.	Measures completed by parents.				<ul style="list-style-type: none"> • Low recruitment rate • Adaptation of SCI for asthma and CF unclear • Means for PSI-SF and SCI not provided • Small sample size for individual illness groups • Age of onset or illness duration not included as confounds.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
3. Britton, 1999 UK	Cross sectional qualitative survey to explore families' experience of JCA from diagnosis to engagement in home programmes.	46 parent-child dyads. Child had primary diagnosis of JCA Children: 72% female. Age range 2.3yrs – 14.08yrs. Recruited from clinic.	Qualitative statements from questions related to impact of JCA on family life & adherence.	Quantitatively determined through questions related to understanding of home treatment & time spent completing different aspects (e.g. splinting & exercise).	Descriptive statistics. Qualitative data entered & coded to create categories with QSR NUDIST 4.0 series software. Specifics of analysis not provided in article.	Adherence influenced by: <ul style="list-style-type: none"> • Pressure on parental time • Desire to reduce conflict with JCA child • Spend time / energy with siblings. JCA caused large amounts of fatigue, stress, grief & conflict. Parents	CASP = 17/20 (strong). Strengths: <ul style="list-style-type: none"> • Understudied disease • Captured a range of family experience • Appropriate data analysis. Limitations: <ul style="list-style-type: none"> • Unknown recruitment rate

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						reported reduced employability.	<ul style="list-style-type: none"> • Gender & age of parents unknown • One recruitment site • Pilot stage • Questions not based on validated measures • Potentially unrepresentative sample with > females.
4. Burgess et al., 2008 Australia	Prospective correlation study.	51 parent – child dyads. Child has asthma and	Parenting Experience Survey developed	1) Percentage of prescribed doses registered by	Correlations in SPSS.	<ul style="list-style-type: none"> • Low adherence (median 70.5%). 	CEBM = 13/24 (weak). Strengths:

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	Examine accuracy of subjective adherence measures in asthma and factors associated with adherence.	taking preventive medicine. Mean age = 3.4 years; range = 18months to 7 years. 31 children were male.	from Living with Children Survey (Tully et al., 1999). Measured after 4 weeks of Smarthinhaler use.	Smarthinhaler, 2) verbal parent judgement of adherence and 3) Physician also estimated adherence. All completed after 4 weeks using Smarthinhaler.	Power calculations determined in PASS.	<ul style="list-style-type: none"> Increasing parental stress led to reduced adherence ($p = 0.05$). Correlation between physician estimated and Smarthinhaler adherence was weak ($R^2 = 0.26$, $p < 0.001$). No relationship between 	<ul style="list-style-type: none"> Prospective study design Operationalised adherence with three measures Included confound of age in correlation between parental stress and adherence. <p>Limitations:</p> <ul style="list-style-type: none"> Small sample

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						parental education or income and adherence. • Trend negative relationship between level of responsibility child had and adherence and trend positive relationship between level of belief in medication and adherence.	• Unclear recruitment method and rate • Non-validated parental stress measure • Level of parental stress not reported.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
5. Celano et al., 2011 USA	Cross- sectional survey. Determine validity of FAMSS (McQuaid, Walders, Kopel, Fritz, & Klinnert, 2005) for low income African- American families.	43 parent- child dyads. Child had asthma and parent needed to be experiencin g parental stress. Parental mean age = 38.6 years (range 25- 69years).	PSI-SF (Abidin, 1990) and BSI (Derogatis, 1993). Complete FRI with parents and RQ with children to assess family functioning.	MDIC to assess inhaler use and review of medical notes to determine healthcare visits (primary care only). Also completed P3C-R to assess parental perception of care.	Cronbach α assess FAMSS internal reliability Correlations assess relationship between FAMSS and others measures or variables (e.g. healthcare visits).	<ul style="list-style-type: none"> High levels of parental stress (mean PSI-SF = 84, SD = 20.6; mean BSI = 57.1, SD = 10.9). N.B. mean PSI-SF high but below clinical cut off of 90. High levels of MDIC errors (67.6%). FAMSS summary score 	CEBM = 17/24 (strong). Strengths <ul style="list-style-type: none"> Thorough analysis Level of parental stress and adherence provided Clear analysis Recruit understudied sample (urban African-American families)

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		86% mothers. Child mean age = 10.5 years, 26 male. 98% of both parents and children African American. Recruited from urban hospital and		Also complete the FAMSS (measure of asthma management).		Cronbach α = 0.87 determined as 'satisfactory'. • PSI-SF negatively correlated with FAMSS summary score ($r=-.41$, $n=43$, $p=.006$). • Parenting stress scores predict asthma management and account for incremental	<ul style="list-style-type: none"> Multiple validated measures. Limitations: <ul style="list-style-type: none"> Cross sectional design Sample size not based on power calculation Due to specificity of sample, results may not be generalisable to other groups.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		1week residential asthma camp (current study a subset from larger RCT).				variance in FAMSS summary score (change in $R^2=.075$, $\beta=-.294$, $SE=.009$, $p=.052$). • FRI positively correlated to FAMSS summary score ($r=.33$, $n=43$, $p=.031$).	
6. Chaney et al., 1989	Cross sectional survey.	25 children with JRA & parents.	FILE (McCubbin &	Adherence to medication assessed by:	Pearson correlation followed by	Mean FILE = 16.6, clinical	CEBM = 15/20 (moderate).

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
USA	Understand impact of family factors on disease management. Circumplex Model of Marital & Family Systems (Olson, Sprenkle, & Russell, 1979) used to determine	Children: 64% female, mean age = 12.5 years Recruited from clinic.	Patterson, 1987). Other family variables included: <ul style="list-style-type: none"> • Adaptability • Cohesion • Coping • Satisfaction 	<ul style="list-style-type: none"> • Global compliance - family estimate of weekly compliance (total doses taken) – divided by number of prescribed doses • Percentage compliance – family record doses taken 	quadratic regression equations where appropriate.	<p>interpretation not provided.</p> <p>Correlations below link to percentage compliance (M = 88%)</p> <p>Negative correlation:</p> <ul style="list-style-type: none"> • Family stress $r(16) = -.61$, $p < .006$ • Cohesion $R = .47$, $F(2, 13) = 4.9$, $p < .03$. 	<p>Strengths:</p> <ul style="list-style-type: none"> • 96% recruitment rate • Families span SES range • Use of model to derive variables (used in research with chronic disease before) • Blind clinician rating of disease activity • Strong analysis • Multiple measures of adherence.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	family factors of interest.			over three weeks with diary -divided by prescribed doses.		Positive correlation: <ul style="list-style-type: none"> Father's satisfaction $r(13)=.61, p<.01$ Mother's coping $r(16)=.49, p<.03$. Regimen knowledge did not impact compliance.	Limitations: <ul style="list-style-type: none"> Cross sectional design One recruitment site Age & gender of parents unknown Specifics of family stress not detailed Clinical interpretation of FILE score not provided Unknown inclusion criteria (e.g. age

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
							range or time since diagnosis). • Small sample size • Low statistical power.
7. Chisholm et al., 2007 UK	Cross- sectional study with interviews and questionnaire . Determine predictors of T1D	65 mother- child dyads. Convenience sample from Scottish diabetes clinic	PSI-SF (Abidin, 1990). Also measured family factors with FES (Moos & Moos, 1994) and CBCL-P (Achenbach, 1991).	Three 20minute interviews with mothers for 24hr recall of adherence. Information gathered on: injection frequency, timings, BGM	ANOVA, <i>t</i> - tests, correlations and stepwise multiple regressions.	Level of adherence depended on which aspect considered • 100% compliance with meals / snacks • 59% compliance with x3 injections/day	CEBM = 18/24 (strong). Strengths: • Reasonable sample size • Good recruitment rate • Confounds considered in

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	adherence in young children.	Child mean age = 6.6 years, SD = 1.7, range 2-8years, 42 male.		frequency, meals, snacks and NMES. Also use devised DKQ to assess diabetes knowledge. Glycaemic control determined treatment outcome.		<ul style="list-style-type: none"> 95% had injection within 1hr window 68% had ≥ 3 BG tests 51% exceeded NMES consumption. Parental stress <ul style="list-style-type: none"> 28% mothers had PSI-SF scores above typical levels Parental stress was not 	analysis (e.g. age and sex) <ul style="list-style-type: none"> Reliable measures. Limitations: <ul style="list-style-type: none"> Cross-sectional design Single recruitment site Sample size not determined with power calculation Possibly unrepresentative sample as

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						<p>correlated with knowledge or adherence measures.</p> <p>Adherence</p> <ul style="list-style-type: none"> • Limited variability • Few significant correlations • Greater NMES intake (indicating poor adherence) correlated to more relationship 	generally good adherence.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						difficulties within family.	
8. DeMore et al., 2005 USA	Cross sectional survey with interview, questionnaire s and objective adherence measure. Assess child behaviour, parenting distress and child routine	45 parent- child dyads from two university asthma clinics. Mean child age = 9.69 years, SD = 1.7, range = 6-12 years. 29 male.	PSI-SF (Abidin, 1990). Also measure child routines with CRI (Sytsma, Kelley, & Wymer, 2001) and parental perception of illness severity with FSI (Fritz et al., 1996).	Percentage of doses administered using dose monitor on inhaler. Also assessed lung function with a spirometry assessment.	Correlations and hierarchical regression.	<ul style="list-style-type: none"> • 67% adherence level. • Mean PSI-SF (difficult child subscale = 52.1; parental distress = 34.6) in normal range. • Parental distress and difficult child subscales of PSI-SF significantly positively correlated with 	CEBM = 15/24 (moderate). Strengths <ul style="list-style-type: none"> • Include mothers and fathers • Validated measure of parental stress • Objective measure of adherence • Thorough analysis • Adherence level similar to other samples.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	in adherence among paediatric asthma.	89% caregivers = mothers, 7% = fathers and 4% = other.				adherence and predicted adherence in regression model, $F(3, 42) = 4.75, p < .01$. • The presence of more embedded routines indicated poorer adherence, but correlation was non-significant.	Limitations • Cross-sectional design • Sample size not based on power calculation • Sample possibly unrepresentative as predominantly white, intact families with low- middle SES). • Adherence measure could have been

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
							improved with electronic device.
9. Eddy et al., 1998 USA	Cross sectional survey. Assess impact of parental & family relationships on adherence.	41 children with CF & mothers. Children: 53% female mean age = 6.7 years; SD = 2.67.	PSI-SF (Abidin, 1990).	Relate to: <ul style="list-style-type: none"> • Medication • Chest exercises • Diet • Vitamins. Rated on questionnaire developed for study by <ul style="list-style-type: none"> • Medical staff 	Pearson correlations.	Marital quality and PSI-SF considered in the 'typical' range (mean PSI- SF = 79.5, clinical cut-off = 90) 14 mothers had 'clinical' levels of parental stress. Significant correlations: <ul style="list-style-type: none"> • Higher parenting stress and 	CEBM = 15/24 (moderate). Strengths: <ul style="list-style-type: none"> • Validated parenting stress measure • Families declining study not different to those included on demographics • Good questionnaire

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	(cohesion and adaptability) & parent-child relationships (parental stress) assessed.			<ul style="list-style-type: none"> Parents. 		<p>parent-child dysfunction relate to lower chest exercise compliance (staff report, $p < .05$)</p> <ul style="list-style-type: none"> Better dyadic adjustment and low parenting stress relate to better dietary compliance (parent report, $p < .05$) Low cohesion related to low 	<p>validity and reliability</p> <ul style="list-style-type: none"> Reported compliance for each domain. <p>Limitations</p> <ul style="list-style-type: none"> Cross-sectional design Small sample size Limited statistical power Age of mothers unknown Possibly unrepresentative

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						<p>compliance for diet (staff report, $p < .01$), exercises (parent $p < .05$ & staff report $p < .01$) & medication (staff report $p < .05$)</p> <ul style="list-style-type: none"> • Low adaptability related to lower compliance with medication (staff report, $p < .05$). 	<p>sample due to 53% recruitment rate</p> <ul style="list-style-type: none"> • Adherence measure developed for study and not validated • Generally, highly compliant families (mean 75%) • One recruitment site.
10. Klok et al., 2014	Grounded Theory	20 parents of children	Parental responses during	Relate to ICS.	Data recorded; verbatim	Identify intentional and unplanned non-adherence.	CASP = 18/20 (strong).

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
Holland	qualitative interviews. Determine barriers to adherence in children with asthma receiving guideline based care.	with asthma. Children: 83% female mean age = 5.9 years. Recruited following observation study where adherence measured for 1 year. Created high ($\geq 75\%$)	qualitative interview.	Electronically measured for 1 year (electronic inhalers recording use. Data uploaded at minimum of four clinic visits over the year).	transcripts generated. Coded in Kwalitan software. Inter-rater reliability for transcript coding was good.	Unplanned non- adherence due to • Issues with child self-managing ICS. Parental factors: • Relational or economic stress – children self- manage ICS younger • Desire to reduce stress caused by giving ICS dose	Strengths • Electronic measure of adherence • Adherence monitored by researcher not involved in clinical care • Include families across adherence spectrum • Information saturation determined sample size.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		and low adherence ($<75\%$) groups. Original study recruited through clinic for families with persistent non- adherence.				Intentional non- adherence relates to concerns of ICS side effects or general beliefs about medication.	Limitations: <ul style="list-style-type: none"> • Age & gender of parents unknown • Single recruitment site • Possibly unrepresentative sample due to low recruitment rate.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
11. McElroy et al., 2007 Uganda	Rapid ethnographic study. Use interviews, FGs and observations. Identify barriers to Ponseti treatment adherence for Clubfoot in Uganda.	Interviews Parents (42, primarily female) Medical staff (38) Information gathered from other sources (e.g. community leaders etc.) but unrelated to this study.	Parent & medical staff responses during study.	Parent & medical staff responses during study.	Interviews transcribed & coded. Thematic analysis completed. Outliers considered.	Parental stress affecting adherence: • Financial stress (treatment & travel costs, loss of earnings & / or sale of livestock to fund treatment) • Stress of neglecting other duties (e.g. care of children, cleaning, cooking, farming)	CASP = 19/20 (strong). Strengths • Triangulation of data collection increasing validity • Culture and language considered throughout • Examining adherence in understudied culture

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		Drawn from districts across Uganda.				<ul style="list-style-type: none"> • Relationship stress - partner not supportive of treatment – financially or emotionally • Length of treatment means stresses are long lasting • Non-compliance referred to as “diagnosis of exclusion” (Farmer, 1997). 	<ul style="list-style-type: none"> • Sites across Uganda • Training for assistants completing data collection and on-going consistency checks • Interview tools piloted and corrected prior to study • Data transcribed and coded on computer

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						<p>Other factors influencing adherence:</p> <ul style="list-style-type: none"> • Poverty • Distance to hospital • Resource availability • Treatment itself • Complex & corrupt health system. 	<ul style="list-style-type: none"> • Sample and study style reflect culture and lifestyle in Uganda • Ways forward considered • Adherent and non-adherent families recruited • Fathers included where possible. <p>Limitations</p> <ul style="list-style-type: none"> • Unknown age of respondents or children

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
							<ul style="list-style-type: none"> • Unknown computer software • Privacy limited during interviews • Some findings specific to Uganda or Clubfoot • Unknown ages of children / time since diagnosis • Relatively small sample size.
12. Njuguna et al., 2015	Cross sectional semi-	75 parents visiting paediatric	Interview responses	Interview responses	Descriptive statistics on:	Stress related reasons for non-adherence:	CEBM = 14/24 (moderate).

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
Kenya	structured interview. To explore experiences of families needing oncology treatment in Kenya.	oncology ward recruited through consecutive sampling. Children age range was 0-14 years 70 % of parents were mothers.	rated on scales.	rated on scales.	<ul style="list-style-type: none"> • socio-demographic • child diagnoses • treatment related experiences (e.g. desire for more information, reasons for non-adherence, use of CAMs). 	<ul style="list-style-type: none"> • Financial pressure of treatment (e.g. loss of earnings, travel, hospital cost) • Multiple demands increased due to cancer (e.g. too busy, no food, work, childcare) • Stress as child may be retained in hospital (until bill paid). 	Strengths <ul style="list-style-type: none"> • Examining adherence in understudied culture • 65% recruitment rate • Two independent interviewers • English and Kiswahili questionnaires • Questions piloted to reduce complexity

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						<p>Examples of other barriers:</p> <ul style="list-style-type: none"> • Accessibility of drugs & treatment • Inadequate transport • Child appears ill or well • Alternative treatment. 	<ul style="list-style-type: none"> • Interviews were private. <p>Limitations</p> <ul style="list-style-type: none"> • Cross-sectional design • One recruitment site • Only descriptive statistics provided • No confidence intervals.
13. Robinson et al., 2016	Cross sectional survey.	257 parent/ child dyads recruited	Parental distress included:	Parent & child self-report of last week with DBRS	Descriptive statistics and Pearson's correlation	PIP frequency mean = 90.5 (SD = 24.6).	<p>CEBM = 18/24 (strong).</p> <p>Strengths</p>

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
USA	Examine parental factors that might relate to adolescent T1D adherence & glycaemic control via parental monitoring.	from two T1D clinics. Children 49% females mean age = 12.84 years; SD = 1.24 Parents 91% mothers. Part of RCT treatment programme to maintain	<i>Depressive symptoms</i> (BDI-II; Beck, Steer, & Brown, 1996). <i>Parenting Stress</i> (PIP; Streisand et al., 2001). Anxiety about hypoglycaemia HFS-P (Cox, Irvine, Gonder-Frederick, Nowacki, & Butterfield, 1987).	(Iannotti, Nansel, et al., 2006). Two DIs relating to 24 hour periods (Holmes et al., 2006) assess number of blood glucose checks. Glycaemic control measured at clinic.	coefficients in SPSS 21. SEM in Mplus 6 software (Muthen & Muthen, 1998-2010) with confirmatory analysis, mediation & overall model fit clearly described	PIP distress mean = 83.7 (SD = 26.6) Parental distress in typical range for parents of child with chronic illness. Model indicated: • Lower parental distress correlate with greater parental self-efficacy • Self-efficacy correlated to	<ul style="list-style-type: none"> • 71% recruitment rate • Large sample • Strong analysis • Two recruitment sites • Questionnaires valid and reliable • Questionnaires completed with researcher • Two types of data collected for certain aspects (e.g. adherence) to improve validity.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
		parent involvement in adolescent care to prevent poor adherence.	Also measure <i>parental self- efficacy</i> for diabetes with SEDSM-P (Iannotti, Schneider, et al., 2006), <i>AP</i> with PSI (Steinber, Lamborn, Darling, Mounts, & Dornbusch, 1994) & <i>Parental</i>		Statistics of full model available in Appendix F.	more AP and parental monitoring • Parental monitoring relates to higher levels of adherence & glycaemic control • Parental monitoring also has direct correlation with greater glycaemic control	Limitations • Cross-sectional design • Possibly unrepresentative sample with 69% white and >80% in or above middle- class SES • PIP not independently entered into SEM.

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
			<i>monitoring</i> with DI relating to 24 hour period (Holmes et al., 2006) & PMDC (Ellis et al., 2008).			<ul style="list-style-type: none"> • Greater self-efficacy also directly correlated with greater adherence. No direct correlation between parental distress & adherence.	
14. Rone-Adams et al., 2004 USA	Cross sectional survey.	66 caregivers from Florida MDA.	QRS-SF (Friedrich, Greenberg, & Crnic, 1983).	Measure designed for study. Caregiver rated how	Linear regression. Multiple stepwise	66% reported some non-compliance Mean QRS = 20, clinical	CEBM = 12/24 (weak). Strengths

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
	Examine relationship between caregiver stress and compliance with home exercise programmes.	Child had to be under 18yrs & have a neuro-muscular disorder.	Contain 4 factors: <ul style="list-style-type: none"> • Parent & family problem • Pessimism • Child character. Physical abilities.	often child was told to complete exercises and how often child completed exercises. Researchers generate a compliance score.	regression (using 4 QRS factors). Correlation.	interpretation not provided. QRS score significantly related to compliance score ($F=4.417$, $p<.039$, $R^2=.065$). Multiple regression and correlation show significant negative correlation between parent & family problems & compliance	<ul style="list-style-type: none"> • Valid parental stress questionnaire • Diagnoses representative of Florida MDA • Appropriate analysis. Limitations <ul style="list-style-type: none"> • Clinical interpretation of QRS not provided • Demographic confounds unknown &

Reference & country	Design & aim	Sample	Parental Stress measure	Adherence measure	Analysis	Findings	CEBM (1-24) / CASP (1-20) score and evaluation
						(F=7.526, $p < .001$, $R^2 = .19$); ($r = .345$, $p < .005$).	therefore unexplored • $\leq 30\%$ recruitment rate due to incomplete questionnaires • Adherence measure not validated • No confidence intervals.

Key: ANOVA – ANalysis Of VAriance; AP – Authoritative Parenting; BDI-II – Beck Depression Inventory, Second Edition; BGM – Blood Glucose Meters; BSI – Brief Symptom Inventory; CASP – Critical Appraisal Skills Programme (maximum score = 20); CBCL-P – Child Behaviour Checklist – Parent form; CEBM – Centre for Evidence Based Management (maximum score = 24); CF – Cystic Fibrosis; CRI – Child Routines Inventory; CVS – Child Vulnerability Scale; DBRS – Diabetes Behavior Rating Scale; DI – Diabetes Interview; DKQ – Diabetes Knowledge Questionnaire; FAMSS – Family Asthma Management System Scale; FES – Family Environment Scale; FGs – Focus Groups; FILE – Family Inventory of Life Events and changes; FIRM – Family Inventory of Resources for Management; FRI – Family Relationship Index;

FSI – Functional Severity Index; HFS-P – Hypoglycemia Fear Survey – Parent; ICS – Inhaled Cortico-Steroids; IDDM – Insulin Dependent Diabetes Mellitus; JCA – Juvenile Chronic Arthritis; JRA – Juvenile Rheumatoid Arthritis; M – Mean age in years; MDA – Muscular Dystrophy Association; MDIC – Metered Dose Inhaler Checklist; MDT – Multi-Disciplinary Team; MNES – Non-Milk Extrinsic Sugars; NUDIST – Non-numerical Unstructured Data Indexing; PASS = Power Analysis and Sample Size software; PIP – Pediatric Inventory for Parents; PMDC – Parental Monitoring of Diabetes Care; PPS – Parent Protection Scale; PSI- Parenting Style Index; PSI-SF – Parenting Stress Index Short Form; P3C-R – Parent’s Perception of Primary Care – Revised; QRS-SF – Questionnaire on Resources and Stress Short Form; RQ – Relatedness Questionnaire; SCI – Self-Care Inventory; SD – of age; SEDSM-P – Self-Efficacy for Diabetes Self-Management – Parents; SES – Socioeconomic Status; SEM = Structural Equation Modelling; SIS – Survey Interview Schedule; SPSS – Statistical Package for Social Sciences; T1D – Type 1 Diabetes.

Measures of Adherence and Parental Stress

Assessments of both parental stress and adherence were always present in the studies, but the impact of parental stress on adherence was not always the primary aim. Additionally, how these factors were assessed differed. In qualitative studies the presence of parental stress was measured through questions concerning: the impact of the child's disorder on the family, or what impacted the parent's ability to complete treatment recommendations. The level of parental stress (e.g. high or low) was not quantified in these instances due to the study design. Within the mixed method papers adherence was measured with a quantitative element (e.g. parental estimates of time spent completing exercises [3], or an electronic recording device [10]). Study 11 however, used interviews, with parents answering questions regarding barriers to adherence.

Quantitative studies used a variety of questionnaires to assess both parental stress and adherence, see Table 3. The Parenting Stress Index-Short Form (PSI-SF; Abidin, 1990) was used in five of the 11 quantitative studies. Adherence measures were more varied, with the most frequently used being family report (4, 6, 7), questionnaires designed for the study (4, 9, 14) or monitoring on an inhaler (4, 5, 8). Although some studies enquired about multiple aspects of adherence (e.g. diet, insulin use, exercise, medication, treatment continuation) only one study utilised different measures of adherence (4).

Table 3

Details of parental stress and adherence measures included in systematic review.

Questionnaire	Domains	Present in study
Parental stress		
FILE (McCubbin & Patterson, 1987)	Intra-family	1, 6
	Marital	
	Pregnancy and childbearing	
	Finance and business	
	Work or family transition	
	Illness and care	
	Family loses	
	Family transitions	
	Family legal violations.	
PSI-SF (Abidin, 1990)	Parental distress	2, 5, 7, 8, 9
	Parent-child dysfunctional interaction	
	Difficult child.	
PIP (Streisand et al., 2001)	Frequency of stressful events	13
	Perceived difficult.	
QRS-SF (Friedrich et al., 1983)	Parent and family problems	14
	Pessimism	
	Child characteristics	

Questionnaire	Domains	Present in study
<hr/>		
Physical incapacitation.		
Parent Experience Survey - developed for study from Living with Children Survey (Tully et al., 1999)	Questions related to: How difficult child's behaviour has been How parent felt about parenting Degree of support from partner.	4
Interview responses rated on scales developed for study	NA	12
<hr/>		
Adherence		
Adherence and IDDM (Hanson et al., 1987)	Adherence to blood glucose and urine testing Diet Treatment of hypoglycaemia.	1
SCI (La Greca, 1992, 2004) developed for diabetes and adapted for asthma and CF	Glucose testing and recording. Administration of insulin Regular meal plan Treatment of hypoglycaemia.	2
Monitoring on inhaler	Electronic monitoring (e.g. SmartInhaler) Measure of dose given through inhaler (5, 8).	4, 5, 8
Family report	Typically estimate percentage of adherence.	4, 6, 7
<hr/>		

Questionnaire	Domains	Present in study
Questionnaire developed for study	Varied across studies.	4, 9, 14
Interview responses rated on scales	NA	12
DBRS (Iannotti, Nansel, et al., 2006)	Frequency of diabetes tasks completed over preceding week.	13
DI (Holmes et al., 2006)	Number of blood glucose checks in 24hr period (other aspects can be chosen).	13

Key: Adherence and IDDM – Adherence and Insulin Dependent Diabetes Mellitus; CF – Cystic Fibrosis; DBRS – Diabetes Behavior Rating Scale; DI – Diabetes Interview; FILE - Family Inventory of Life Events; PIP – Pediatric Interview for Parents; PSI-SF – Parenting Stress Index; QRS-SF – Questionnaire on Resource and Stress – Short Form; SCI – Self-Care Inventory;

Adherence Findings

Qualitative studies and one quantitative study did not report adherence levels (3, 10, 11, 12). Most quantitative studies provided adherence levels as a percentage (2, 4, 5, 6, 7, 8, 9, 14), which ranged from 12% for diet (9) to 100% for meals (7). One study provided a mean value for adherence, but no interpretation of whether this was high or low (1). The remaining quantitative study provided a mean for the adherence questionnaire used and described findings as low but normative (13).

Parental Stress Findings

Qualitative findings regarding components of parental stress can be found in Table 4. In brief, these included financial concerns, minimising

conflict with their child, managing time pressures and competing demands, concerns about prognosis and parental relationship difficulties.

Table 4

Aspects related to parental stress in qualitative studies.

Parental stress domain	Related factors	Factor present in study
Financial stress	Travel to treatment centres:	
	1) leading to loss of earnings	11
	2) travel cost.	11
	Treatment cost.	11
	Loss of earnings as parents report becoming less employable.	3
	'Economic' (not defined).	10
Conflict with child	Completing treatment without causing conflict with child.	3, 10
Managing time pressures as parents have additional demands.	Other children	3, 11

Parental stress domain	Related factors	Factor present in study
	Housework	11
	Work	3, 11
	Getting children to school on time	10
	Fitting treatment in around family routine.	3
Fatigue		3
Emotional stress	Concerns for child and prognosis.	11
Parental relationship strains	Long term and additional demands on family	11
	Difference in paternal prioritisation of finances	11
	'Relational' (undefined).	10

In study 11 financial strains were particularly acute and despite a reported desire of parents for their child to be well and function ably in the community they simply could not afford treatment. Therefore, although financial strains related to travel and treatment may seem more logistical or practical, the financial stress led to long-lasting and difficult decisions around adhering to treatment plans.

Quantitative studies did not provide specifics of what constituted parental stress, however, questionnaires included domains such as: marital

strain, parent-child conflict and financial difficulties (Table 3). Three studies did not provide a level of parental stress, these used a questionnaire developed for the study (4), the PSI-SF (2) or scaled interview responses (12). Across the review, the PSI-SF was the most frequently used scale (2, 5, 7, 8, 9) and a cut-off of 90 indicates 'clinical' levels of parental stress (Abidin, 1990). Three studies provided a mean PSI-SF score and 'clinical' levels of parental stress was not indicated in any study (5, 8, and 9). However, these studies interpreted the scores as 'high' but 'normative' for parents whose child has a chronic illness. Only study 7 (no mean provided) and 9 indicated approximately a third of their samples had 'clinical' levels of parental stress (28% and 34% respectively). Study 13 used the Pediatric Inventory for Parents (PIP) to measure parental stress, reporting a mean frequency of parental stress, which was 'normative' for parents of children with a chronic illness. The mean Questionnaire on Resources and Stress (QRS; Freidrich et al., 1983) score in study 14 was found to be higher than the mean provided during the questionnaire's development. However, it remains unclear whether there is a clinical 'cut-off' to indicate if this is significantly different to parents of children without a chronic illness. Finally study 1 provided the clinical interpretation of 'normal' for parents of chronically ill children, but no mean.

Findings Related to Review Aim: Parental stress and Adherence

All qualitative studies found parental stress impacted adherence levels (3, 10, 11). Quantitative studies measured the relationship between parental stress and adherence using correlations (1, 2, 4, 5, 6, 7, 8, 9, 13, 14), regressions (1, 2, 6, 7, 8, 14) and Structural Equation Modelling (SEM; 13).

Six quantitative studies found a direct negative relationship between parental stress and adherence, where greater parental distress significantly negatively affected adherence (2, 4, 5, 6, 8, 14). Two studies found an indirect negative relationship between parental stress and adherence (1, 13). This relationship was mediated by parental satisfaction with care (1), parental self-efficacy in managing diabetes, authoritative parenting (AP) and parental monitoring (13). One quantitative study provided no statistics on the relationship between parental stress and adherence (12). The final quantitative papers either found a positive relationship between parental stress and adherence (8) or no significant relationship (7). Possible explanations for these differences are explored in the discussion. Due to the variability of disorders present in this review, there was no relationship between the pattern of results and the type of child's chronic disorder.

The majority of studies included in this review were of moderate or strong quality. However, the two 'weak' studies (4, 14) found increased parental stress reduces adherence, in line with the main review finding. Therefore, the inclusion of these two studies does not alter the overall finding regarding the relationship between parental stress and adherence.

Additional Factors Found to Affect Adherence

Although this review focussed on understanding the link between parental stress and adherence, additional factors were also identified among the papers included in this review. Supplementary factors found to be associated with adherence were: parental satisfaction with care (1), community stressors (1), family cohesion, parental satisfaction and coping (6),

relationship difficulties (8), parent-child conflict, family cohesion and adaptability (9), health beliefs (10, 12), difficulties in establishing a routine (10), parental education (12), communication with doctors (1, 12), complementary alternative medicines (12), social support (12), self-efficacy in managing disorder (13), authoritarian parenting (13) and parental monitoring (13). Of note, non-significant relationships were also found between parental education or income and adherence (4) and a trend negative correlation was found between using child routines and poor adherence (8).

Discussion

This review identified fourteen studies that examined the relationship between parental stress among parents or caregivers of children with a chronic illness and their child's adherence to treatment. Three were qualitative and eleven were quantitative cross-sectional surveys. Twelve studies had 'moderate' or 'strong' quality ratings. Two quantitative studies scored as 'weak' due to reporting omissions on recruitment.

Parental Stress and Adherence

Across included studies many factors contributed to parental stress in the context of caring for a child with a chronic illness. These included, but were not limited to, financial stress, time pressures, relationship and emotional strains. Parental stress is therefore a multifaceted concept. It is notable therefore that, overall the review found a negative relationship between parental stress and children's adherence. In qualitative studies this was implicated through parental interview responses. Within quantitative studies, eight found either a direct or an indirect negative relationship where

increasing parental stress correlated with poorer adherence. The presence of such a relationship, given the multifaceted nature of parental stress, demonstrates the utility of considering it as one concept. Studies not reporting this relationship, or where the negative relationship was mediated by additional factors, will now be considered.

The three quantitative studies that did not report a negative relationship between parental stress and adherence either only reported descriptive statistics (12) or found no (7) or a positive relationship (8) between parental stress and adherence. Whilst it seems these discrepancies reduce the strength of the overall review finding, when individual studies are considered, this may not be the case. In relation to study 12, the study aim was answered sufficiently through descriptive statistics. It is possible therefore that a relationship between the accounts of parental stress and poor adherence reported could have been found with thorough statistical analysis. The absence of a relationship in study 7 may reflect the high levels of adherence within the sample. To determine these factors, study replication using thorough statistical analysis and a more representative sample is required. Finally, study 8 found high levels of difficult behaviour among the children in their sample. Authors hypothesised this led to the increased levels of parental monitoring found and in turn increased adherence. Therefore, the positive relationship between parental stress and adherence may reflect a specific aspect of parental stress related to fear of harm coming to their child, which resulted in increased investment in adherence to improve their child's outcomes. Although it is not possible to determine the exact reason for this positive relationship, study 8 demonstrates the importance of considering

parental stress to enhance understanding of all ways it may impact adherence.

Two studies noted an indirect relationship between parental stress and adherence (1, 13). Study 1 found mothers who experienced higher levels of stress were less satisfied with medical care (including access, cost and doctor's manner), which in turn was related to poorer adherence. The authors were interested in whether parental satisfaction impacted adherence and therefore did not directly examine the relationship between parental stress and adherence. However, as this study found that mothers with higher parental satisfaction had lower levels of parental stress, a direct relationship between parental stress and adherence may have been present. Notably though, this study demonstrates how system resources and professionals can impact parental stress levels. Study 13 was the only study to complete SEM and found that parental stress may lead to poor adherence through the impact of stress on parenting capacities (e.g. maintaining greater levels of parental self-efficacy, AP and parental monitoring). Further studies considering causality in this way, however, are needed.

The five studies that either did not find a negative relationship or reported mediating factors therefore do not detract from the overall review finding that increased parental stress is typically related to poorer levels of adherence among children with a chronic illness. Instead these studies demonstrate the need for replication of studies with thorough analysis (12) and representative samples (7) as well as the complexity of parental stress and its relationship to adherence (1, 8, 13).

The presence of a relatively consistent medium-large negative relationship between parental stress and adherence was apparent across a range of paediatric chronic illness. Therefore, this review supports suggestions previous reviews (Cousino & Hazan, 2013; Morawska, Calam, & Fraser, 2015; Psihogios et al., 2019) to collate data across disorders to enhance understanding of a particular factor, such as parental stress.

Strengths and Limitations

A main strength of this review is its systematic nature. Dutifully subscribing to clear guidance enabled this current paper to systematically search, identify and review relevant literature to reduce bias as far as possible. This included for example, consulting experts on the identification of key terms, naming PICOS criteria and ensuring papers were independently second rated at two separate stages. Despite these methodological strengths, a future review would be advanced through the inclusion of unpublished literature.

The inclusion of critical appraisal tools was also a strength of this review. Guidance was followed to support the identification of appropriate tools. However, these tools would be further enhanced through the inclusion of scoring and interpretation recommendations. Currently, the tools provide information to enable item coding; however, no structure is provided to ensure that 'strong', 'moderate' and 'weak' papers are being coded similarly across reviewers. This limitation was avoided within the current review through the inclusion of a second rater. However, the assessment of future reviews utilising these tools would be improved with more detailed and consistent guidance on how to interpret the outcomes of the study quality ratings.

Reviewing the concept of parental stress in relation to adherence across paediatric chronic health conditions was also a strength of this review. Adherence had been identified as potentially being improved via interventions to address parental stress (Cousino and Hazen, 2013). However, no review had sought to better understand the relationship between parental stress and adherence for children with long-term health conditions. This review therefore advances understanding in this field. The clinical importance of this is discussed in the following section.

Clinical Importance and Future Research

This is the first review to assess the relationship between parental stress and adherence among children with a chronic illness irrespective of diagnosis. Improving understanding regarding adherence through thinking trans-diagnostically is a relatively recent change. Cousino & Hazen (2013) and Psihogios et al., (2019) demonstrated the utility of this work; however, neither assessed the impact of parental stress on adherence. Parental stress is important to consider due to the well-recognised low levels of adherence among paediatric chronic conditions and the unique role parents play in adherence for children. Parental stress has also been suggested as a potential target for interventions to improve adherence (Cousino & Hazan, 2013). Improving adherence is a global issue and may be more impactful than developing new treatments for chronic conditions (WHO, 2003). Reviews, such as the current one, are therefore critical to ensure future interventions to improve adherence are evidence based.

In relation to the pediatric self-management framework (Modi et al., 2015), the findings from this review support the identification of modifiable

processes to improve adherence and in particular, parental stress, as suggested by Cousino and Hazan (2013). However, the multifaceted nature of parental stress suggests that particular aspects are more amenable to intervention at different levels (e.g. governmental policy, clinical practice or individualised care). For example, interventions to reduce parental stress via reducing the financial pressures, may target charities or government policy. Charities could be encouraged to make clinics more aware of the support they provide to ensure improved signposting. Government policy amendments, however, may be more appropriate for a larger scale measure (e.g. tax relief or increased access to benefits). In contrast, if parental stress is being targeted through enabling families to better manage possible treatment related conflicts, then a more patient-centred approach will be needed. Morawska et al. (2015) conclude that such interventions should include aspects of psychoeducation and concrete strategies for families. Psychoeducation should relate to the illness and highlight the association between illness management and child adjustment. In turn, strategies should support parents to create and embed illness management within the home (e.g. consistent routines) and to help their child with potential future anxiety or social, emotional or behavioural difficulties. Kaminski et al. (2008) highlight that interventions should focus on creating positive interactions between parent and child as well as enabling consistency. A specific parenting intervention to reduce parental stress (and as such improve adherence) could include psychoeducation and strategies as well as supporting the development of positive parenting interactions. Components of psychoeducation and parenting strategies may need to be tailored to specific

disorders; however, interventions could and should be rolled out across disorders (Morawska et al., 2015).

Findings from this review also identified reporting heterogeneity for parental stress. Overall, the studies typically reported 'high' but normative levels of parental stress for parents of children with a chronic illness, supporting Cousino & Hazen (2013). However, due to the variety of measures used, a threshold score indicating 'high' levels of parental stress was not clear. This review found the PSI-SF was the most frequently cited measure of parental stress. However, the PSI-SF was not designed to specifically measure parental stress among parents whose child is chronically ill. Cousino & Hazen (2013) found PIP (designed specifically for this population), was the most cited tool; however, only study 13 utilised this in this review. No clinical cut-off is available for the PIP. Future research would benefit from using a standard measure of parental stress and consistently reporting results related to the level of parental stress and a clinical interpretation.

Clinically, the finding of high but normative levels of parental stress within this population indicates parental stress is prevalent among parents of children with a chronic illness, regardless of the child's diagnosis. Findings suggest a cumulative impact on adherence – with increasing levels leading to poorer adherence. However, research is required to help clinical practice identify a possible cut-off that indicates a significant impact on adherence to enable efficient support targeting. However, clinics could introduce a parental stress screening into practice to enable families to start discussing this topic. In time this could aid understanding about a 'clinical' score.

The medium-large negative relationship between parental stress and adherence was notable across disorders and countries. Study 9 ('moderate' quality) was the only study to report a negative relationship of small effect. All others (1, 2, 4, 5, 6, 13, 14) reported medium to large effect sizes. In study 8, a positive relationship of medium effect was found. Therefore, despite the heterogeneity present in this review, it was possible to collate results and enhance understanding regarding barriers to adherence. Two studies from Africa (Uganda and Kenya) were included, demonstrating the global importance of adherence, in line with the WHO report (WHO, 2003). However, the majority of research was completed within western cultures and no studies from Asia or South America were included. Future research will continue to benefit from thinking trans-diagnostically and globally.

Finally, this review identified additional relationships to adherence, such as health beliefs, parenting style and family communication. Studies excluded at screening often investigated similar factors. Whilst not within this review's scope to consider these factors, it is clear factors beyond parental stress may affect a child's adherence. Reviews aiming to understand whether these factors also influence adherence across paediatric chronic health conditions and to what extent, would be a welcome addition to the field. Additionally, the nature of how these factors interact with each other to influence adherence (or outcomes such as child health) is likely to be complex, as demonstrated by study 14. Therefore, research could attempt to combine understanding on the relationship between individual factors and adherence.

Conclusion

This is the first review to consider the relationship between parental stress and adherence for children with a chronic illness. Findings note the complexity of parental stress as a concept as it contains aspects such as time pressure, relationship factors and financial pressures. However, the review typically found parents had high levels of parental stress and that increasing levels of parental stress are related to poorer levels of child adherence. Parental stress is therefore one aspect that could be targeted by interventions to improve adherence. The intervention design will depend on the aspect of parental stress and population being targeted. The review also identified additional factors that impact adherence (e.g. health beliefs), which deserve further research.

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Appendix A: Mapped Terms for each Database

Table 5

Mapped terms for each database.

Database	MEdical Subject Headings	Mapped terms
PsycINFO	Child	Child behavior Child psychology Father child communication Father child relations Mother child communication Mother child relations Parent child communication Parent child relations.
	Adolescent	Adolescent psychiatry Adolescent behaviour Adolescent attitudes Adolescent development Adolescent psychology Adolescent health.
	Pediatric	Pediatrics Parents Childhood development.
	Chronic disease	Chronic illness Chronicity (disorders) Disease management Diabetes Health behavior Self management Health Health promotion Quality of life.
	Parental stress	Stress Parents Autism spectrum disorders Behavior problems Parent child relations Parenting Parental attitudes Social support Mothers Coping behaviour Parental characteristics Family relations Caregiver burden Socioeconomic status Adjustment.

Database	MEdical Subject Headings	Mapped terms
	Adherence	Health behaviour Neuroleptic drugs Antiviral drugs Treatment outcomes Diabetes HIV Chronic illness Hypertension Drug therapy Intervention Treatment compliance Compliance Client attitudes Exercise.
	Prescription medication	Epidemiology Advertising Drug usage Risk factors Side effects (drugs) Drug abuse Major depression College students Drug therapy Treatment compliance Prescription drugs Client attitudes Geriatric patients Opiates.
	Non-adherence	Drug therapy Treatment compliance Prescription drugs Client attitudes.
	Health behavior	Family Health care policy Knowledge level Health promotion Behaviour change Health Human development Behaviour Technology Health behaviour Experimentation Diets.
Medline	Child	Child Child behavior

Database	MEdical Subject Headings	Mapped terms
		Child behavior disorders Child development Child health Child health services Child hospitalized Child nutrition disorders Child, preschool Child psychiatry Father child relations Mother child relation Only child Parent child relations Child development disorders, pervasive Child psychology Child nutrition sciences.
	Pediatrics	Child Adolescent Child, preschool Infant Pediatrics HIV infections.
	Adolescent	Adolescent medicine Adolescent behaviour Adolescent Adolescent health Adolescent psychiatry Adolescent development Psychology, adolescent.
	Chronic disease Parental stress	Chronic disease Child Child, preschool Stress, psychological Parenting Adolescent Parents Adult.

	Adherence	medication adherence guideline adherence treatment adherence and compliance.
	Prescription medication	Prescription drugs Drug prescriptions.
	Health behavior	Health behaviour.
EMBASE	Child	Handicapped child Child health care Preschool child Child psychology Child Child psychiatry Child development Child behavior Child parent relation School child Child health Mother child relation Father child relation.
	Pediatrics	pediatrics
	Adolescent	Adolescent behavior Adolescent health Adolescent Adolescent development Adolescent family inventory of life events and changes Adolescent depression Adolescent disease.
	Chronic disease	Chronic disease.
	Parental stress	Child parent relation Child Parent Parental stress Adult Stress.
	Non-adherence	Patient Patient compliance Drug therapy Medication compliance.
	Prescription medication	Prescription Patient Drug therapy.
	Health behavior	Preventative medicine Attitude to health Behavior Health behavior

		Health education Prevention.
CINAHL	Child	Child Child, Disabled Child Behavior Parents of Disabled Children Adult-Child Relations Child Health Child Psychology Child Psychiatry Child, Preschool Parent-Child Relations Mother-Child Relations Child Behavior Alteration (Saba CCC) Father-Child Relations Child Development Child Development: Middle Childhood (6-11 Years) (Iowa NOC) Child Development: Adolescence (12-17 Years) (Iowa NOC) Child Development: 6 Months (Iowa NOC) Child Development: 5 Years (Iowa NOC) Child Development: 12 Months (Iowa NOC) Child Development: 4 Months (Iowa NOC) Child Development: 3 Years (Iowa NOC) Child Development: 2 Years (Iowa NOC) Child Development: 2 Months (Iowa NOC) Child Development: 4 Years (Iowa NOC) Child Adaptation to Hospitalization (Iowa NOC) Psychology, Developmental Pediatrics Parents Parenting Alteration (Saba CCC) Parenting (Iowa NOC) Parenting Infant Development Infant Behavior Behavior.
	Pediatric	Hospitals, pediatric Pediatric units Rehabilitation, pediatric Pediatric care+ Pediatrics+ Child psychology Rehabilitation+.
	Adolescent	Adolescent behavior Adolescent psychology Adolescent psychiatry Adolescent medicine Adolescent health Adolescent development

	Adolescence+
	Child development: adolescence
	Adolescence+
	Child development: Adolescence (12-17years)(IOWA NOC)
	Rehabilitation, pediatric
	Rehabilitation+
	Parenting
	Parents+
	Health+
	Child psychology
	Child psychiatry
	Child health
	Child development
	Child behavior+
	Child+
	Behavior+
	Adult-child relations.
Chronic disease	Chronic disease+
	Infant, newborn, diseases+
	Attitude to illness+
	Psychosocial aspects of illness+
	Disease+
	Treatment behavior: illness or injury (IOWA NOC)
	Disease management+
	Self-management
	Rehabilitation+
	Pediatrics+
	Noncompliance of therapeutic regimen (Saba CCC)
	Intervention trials
	Ineffective management of therapeutic regimen: Families (NANDA), individuals (NANDA), Community (NANDA)
	Health psychology
	Disability management
	Coping strategies questionnaire
	Coping health inventory for parents
	Adolescent-family inventory of life events and changes.
Parental stress	Parental behavior
	Parents of disabled children
	Parental attitudes+
	Stress, physiological+
	Parenting education
	Parenting (IOWA NOC)
	Parenting
	Caretaking-parenting (Omaha)
	Parent-infant relations+

	Parent-child relations+
	Stress, psychological
	Role stress
	Caregiver burden
	Parents+
	Parental role+
	Stress management
	Parenthood+
	Coping health inventory for parents
	Stress+
	Life change events+
	Compassion fatigue
	Paternal attitudes
	Mothers+
	Fathers
	Adult-child relations.
Adherence	Guideline adherence
	Adherence behaviour (IOWA NOC)
	Medication compliance
	Patient compliance+
	Noncompliance (NANDA)
	Compliance with therapeutic regimen (Saba CCC)
	Compliance with medication regimen (Saba CCC)
	Compliance with medical regimen (Saba CCC)
	Compliance care (Saba CCC)
	Compliance with diet (Saba CCC).
Prescription medication	Prescriptions, non-drug
	Prescriptions, drug+
	Attitude to medical treatment
	Self-medication
	Alternative therapies+
	Prescription drug monitoring programs
	Drugs, prescription
	Drugs, non-prescription
	Medication management
	Medicine+
	Medication treatment (Saba CCC)
	Medication regimen (Omaha)
	Medication care (saba CCC)
	Noncompliance with medication regimen (Saba CCC)
	Compliance with medication and medical regimen (Saba CCC)
	Medication management (Iowa NIC)
	treatment refusal
	Self administration+
	Psychosocial adjustment to illness scale

	Patient compliance
	Patient autonomy
	Drug utilization+
	Decision support techniques+.
Health behavior	Health behavior+
	Health seeking behavior alteration (Saba CCC)
	Health behavior component (Saba CCC)+
	Cox interaction model of client health behavior
	Health seeking behavior (Iowa NOC)
	Health promoting behavior (Iowa NOC)
	Health knowledge and behavior (Iowa NOC)+
	Health behavior (Iowa NOC)+
	Health seeking behaviors (NANDA)+
	Domain IV: Health-related behaviors domain (Omaha)+
	Knowledge: Health behaviors (Iowa NOC)
	Paternal behavior
	Maternal behavior
	Behavior modification+
	Infant behavior
	Child behavior+
	Behavioral changes
	Help seeking behavior
	Adolescent behavior
	Parental behavior
	Behavior modification (Iowa NIC)
	Behavior management (Iowa NIC)
	health beliefs
	Child health
	Attitude to health+
	Outcomes (health care)+
	Infant behavior alteration (Saba CCC)
	Child behavior alteration (Saba CCC)
	Adult Behavior Alteration (Saba CCC)
	Adolescent Behavior Alteration (Saba CCC)
	Maternal-Child Health
	Health Psychology
	Health Maintenance Alteration (Saba CCC)+
	Health Belief Model
	Adolescent Health
	Infant Behavior (NANDA)+
	Compliance Behavior (Iowa NOC)
	Adherence Behavior (Iowa NOC)
	Transtheoretical Stages of Change Model
	Health Beliefs (Iowa NOC)

Health Beliefs: Perceived Threat (Iowa
NOC)
Health Beliefs: Perceived Resources (Iowa
NOC)
Health Beliefs: Perceived Control (Iowa
NOC)
Health Beliefs: Perceived Ability to Perform
(Iowa NOC)
Health Belief (Iowa NOC)+
Health and Life Quality (Iowa NOC)+
Health and Disease+
Coping Health Inventory for Parents
Caregiver Physical Health (Iowa NOC)
Caregiver Emotional Health (Iowa NOC)
Treatment Refusal
Self Regulation
Refusal to Participate
Refusal to Treat
Patient Preference
Patient Attitudes
Parenting
Parental Attitudes+
Life Style Changes
Ineffective Management of Therapeutic
Regimen: Families (NANDA) Ineffective
Management of Therapeutic Regimen:
Individuals (NANDA)
Ineffective Management of Therapeutic
Regimen: Community (NANDA) Ineffective
Individual Coping (NANDA)
Ineffective Family Coping, Disabling
(NANDA)
Ineffective Family Coping, Compromised
(NANDA)
Guideline Adherence Effective
Management of Therapeutic Regimen:
Individual (NANDA)
Disability Management
Caretaking-Parenting (Omaha)
Caregiver Well-Being (Iowa NOC)
Caregiver Support (Iowa NIC)
Caregiver Performance: Direct Care (Iowa
NOC)
Coping+
Attitude to Illness+
Attitude Measures
Adolescent Medicine.

Appendix B: Qualitative Critical Appraisal Tool



Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- what was the goal of the research
- why it was thought important
- its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
- Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

4. Was the recruitment strategy appropriate to the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the researcher has explained how the participants were selected
 - If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
 - If there are any discussions around recruitment (e.g. why some people chose not to take part)

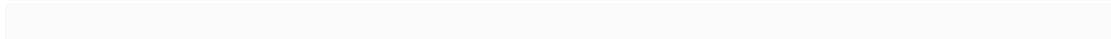
Comments:

5. Was the data collected in a way that addressed the research issue?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the setting for the data collection was justified
 - If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
 - If the researcher has justified the methods chosen
 - If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
 - If methods were modified during the study. If so, has the researcher explained how and why
 - If the form of data is clear (e.g. tape recordings, video material, notes etc.)
 - If the researcher has discussed saturation of data

Comments:



6. Has the relationship between researcher and participants been adequately considered?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
 - How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
 - If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
 - If approval has been sought from the ethics committee

Comments:

8. Was the data analysis sufficiently rigorous?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If there is an in-depth description of the analysis process
 - If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
 - Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
 - If sufficient data are presented to support the findings
 - To what extent contradictory data are taken into account
 - Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

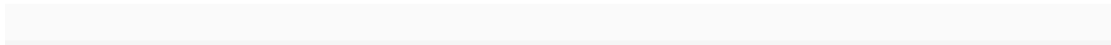
Comments:

9. Is there a clear statement of findings?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

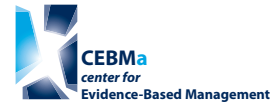
- HINT: Consider whether
- If the findings are explicit
 - If there is adequate discussion of the evidence both for and against the researcher's arguments
 - If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
 - If the findings are discussed in relation to the original research question

Comments:



Section C: Will the results help locally?	
10. How valuable is the research?	<div>HINT: Consider<ul style="list-style-type: none">• If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature• If they identify new areas where research is necessary• If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used</div>
<div>Comments:</div>	

Appendix C: Quantitative Critical Appraisal Tool



Critical Appraisal of a Survey

Appraisal questions	Yes	Can't tell	No
1. Did the study address a clearly focused question / issue?			
2. Is the research method (study design) appropriate for answering the research question?			
3. Is the method of selection of the subjects (employees, teams, divisions, organizations) clearly described?			
4. Could the way the sample was obtained introduce (selection) bias?			
5. Was the sample of subjects representative with regard to the population to which the findings will be referred?			
6. Was the sample size based on pre-study considerations of statistical power?			
7. Was a satisfactory response rate achieved?			
8. Are the measurements (questionnaires) likely to be valid and reliable?			
9. Was the statistical significance assessed?			
10. Are confidence intervals given for the main results?			
11. Could there be confounding factors that haven't been accounted for?			
12. Can the results be applied to your organization?			

Adapted from Crombie, *The Pocket Guide to Critical Appraisal*; the critical appraisal approach used by the Oxford Centre for Evidence Medicine, checklists of the Dutch Cochrane Centre, BMJ editor's checklists and the checklists of the EPPI Centre.

Appendix D: Quality Rating for Quantitative Studies

Table 6

Detailed CEBM scores for quantitative studies included in review.

Study	Aim	Design	Selectio	Bias*	Representativ	Powe	Recruitme	Measurement	Questions				Total
									Statistical	CIs	Confounds	Relevanc	
			n		e	r	nt	s	significanc		*	e	
									e				
1	2	2	2	2	1	0	1	2	2	0	1	2	17
2	2	2	2	2	1	0	0	2	2	0	0	2	15
4	2	2	0	1	1	2	1	1	2	0	0	1	13
5	2	2	2	2	2	0	2	2	2	0	0	1	17
6	2	2	1	1	1	0	2	1	2	0	1	2	15
7	2	2	2	2	1	0	2	2	2	0	2	1	18
8	2	2	2	1	1	0	2	2	2	0	0	1	15
9	2	2	2	1	2	0	0	2	2	0	1	1	15

PARENTAL STRESS AND CHILD ADHERENCE										113			
13	2	2	2	2	2	0	2	0	0	NA	2	0	14
14	2	2	2	2	2	0	2	2	2	0	1	2	18
15	2	2	2	0	0	0	0	2	2	0	0	2	12

Key; CEBM – Centre for Evidence Based Management; CIs – Confidence Intervals. For most items 2 = ‘yes’, 1 = ‘CT’, 0 = ‘no’. For
 * items the scores are reverses – ‘yes’ = 0, ‘CT’ = 1 and ‘no’ = 2.

Appendix E: Quality Rating for Qualitative Studies

Table 7

Detailed CASP scores for included qualitative studies.

Questions											
Study	Section A: Validity					Section B: Results				Section C: Helpful	
	Aims	Methodolog y	Design	Recruitmen t	Data	Researche r	Ethics	Analysi s	Finding s	Value	Total
2	2	2	2	2	2	2	1	0	2	2	17
10	2	2	2	2	2	2	2	1	1	2	18
11	2	2	2	2	2	2	2	1	2	2	19
12	2	2	2	1	2	0	2	2	2	2	17

Key: CASP – Critical Appraisal Skills Programme. Scored where ‘yes’ = 2, ‘CT’ = 1, ‘no’ = 0.

Appendix F

Figure 2

Full structural equation model from Robinson et al. (2016).

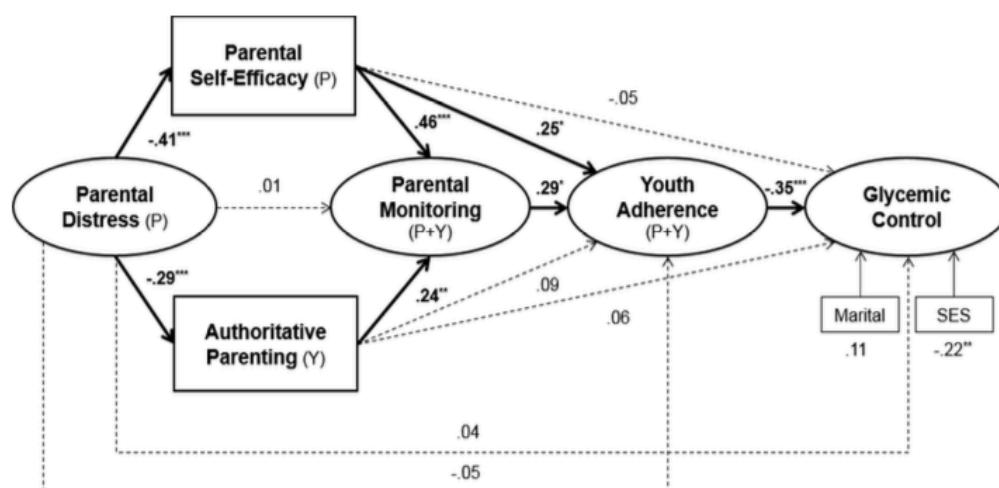


Figure 2.

Final structural equation model. Values shown are standardized regression coefficients.

Source of data for each construct is provided within parentheses: P=Parent, Y=Youth, P

+Y=Parent and Youth report. * $p < .05$, ** $p < .01$, *** $p < .001$

Appendix G: Dissemination Statement and Author Instructions

Dissemination Statement

This paper will be submitted to the Journal of Pediatric Psychology for publication.

Instructions to authors

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including title page, references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Review articles should not exceed 30 pages. Invited commentaries should be discussed with the Editor. The *Journal of Pediatric Psychology* no longer accepts brief reports but will accept manuscripts that are shorter in length.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (6th ed.). There are two exceptions:

The academic degrees of authors should be placed on the title page following their names, and a structured abstract of not more than 250 words should be included. The abstract should include the following parts:

1. Objective (brief statement of the purpose of the study);
2. Methods (summary of the participants, design, measures, procedure);
3. Results (the primary findings of this work); and
4. Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch).

Informed consent and ethical treatment of study participants: Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Review articles:

(a) *Topical Reviews*: Topical reviews summarize contemporary findings, suggest new conceptual models, or highlight noteworthy or controversial issues in pediatric psychology. Topical reviews are not intended to provide short data summaries or syntheses. Rather they are intended to foster new ways of thinking about a topic area and provide a direction for future research or practice. They are limited to 2,000 words, contain no more than 2 tables or figures, and have an upper limit of 30 references. Supplementary online material (e.g., additional tables) may be considered on a case by case basis.

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Please consult this editorial ([New Guidelines for Publishing Review Articles in JPP](#)) which further describes guidelines for review articles, and the Checklist for Preparing and Evaluating Review Articles.

Additional Guidance

The following links provide additional guidance for authors and reviewers: Editorial Policy, Authors' Checklist, Guidelines for Reviews, Suggestions for Mentored Reviews, "People First," NIH policy, Replication of research, Duplicate and redundant policies, Conflict of interest.

See the following articles for detailed guidance concerning preparation of manuscripts: Editorial: Thoughts in Improving the Quality of Manuscripts Submitted to the *Journal of Pediatric Psychology*; How to Write a Convincing Introduction; Methods: Editorial: How to Report Methods in the *Journal of Pediatric Psychology*; Results and Discussion: Editorial: How to Write an Effective Results and Discussion Section for the *Journal of Pediatric Psychology*.

Funding

Details of all funding sources for the work in question should be given in a separate section entitled "Funding." This should appear before the "Acknowledgements" section.

The following rules should be followed:

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- The full official funding agency name should be given, i.e. "the National Cancer Institute at the National Institutes of Health" or simply "National Institutes of Health," not "NCI" (one of the 27 subinstitutions) or "NCI at NIH" (full RIN-approved list of UK funding agencies)
- Grant numbers should be complete and accurate and provided in parentheses as follows: "(grant number xxxx)"
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SCHOOL OF PSYCHOLOGY
DOCTORATE IN CLINICAL PSYCHOLOGY

EMPIRICAL PAPER

**A distance-based intervention supporting neuropsychological
recommendations for children with a neurodisability**

Trainee Name:	Dr Jessica Watts
Primary Research Supervisor:	Dr Jennifer Limond Senior Lecturer & Consultant Clinical Neuropsychologist
Secondary Research Supervisor:	Dr Anke Karl Senior Lecturer
Target Journal:	The Clinical Neuropsychologist
Word Count:	7,966 words (excluding abstract, figures, references, appendices)

**Submitted in partial fulfilment of requirements for the Doctorate Degree in Clinical
Psychology, University of Exeter**

Abstract

Background: Only 50% of paediatric neuropsychological recommendations are adhered to because parents struggle to understand them and can feel too overwhelmed to implement them. The Information-Motivation-Behaviour skills (IMB) model offers a novel way to design neuropsychological interventions that specifically address parental barriers and thus improve adherence.

Method: Four families, recruited through a national clinic, completed a single-case experimental multiple baseline design study. An IMB-informed intervention comprising a baseline and intervention phase was designed to improve adherence to neuropsychological recommendations for children with rare neurological conditions. A pre-baseline measure of recommendation implementation informed by conversations with families was completed retrospectively. An online daily outcome measure regarding the implementation of eight recommendations was completed over thirty-one days. Three (of the eight) recommendations, 'targeted recommendations,' were chosen by families in collaboration with the researcher as the focus for the intervention. The remaining five were 'non-targeted recommendations.' Visual analysis, randomisation tests and effect size tests of adherence were completed. Pre and post intervention measures of parental stress and child's everyday functioning were completed and analysed using the Reliable Change Index (RCI). Parents provided feedback regarding the acceptability of the intervention.

Results: Two children in the study had diagnoses of Sturge-Weber Syndrome (SWS), one with Congenital Melanocytic Nevus (CMN) and one had a chromosomal disorder. Implementation of targeted recommendations significantly increased

following the intervention when data was analysed as a group. No significant increases were found for non-targeted recommendations. Visual analysis did show implementation increases from pre-baseline to baseline and for non-targeted recommendations, suggesting the simple information and focusing of recommendations provided at study commencement enabled some improved adherence. However, these increases were typically not sustained and were not as large as increases following the intervention for targeted recommendations. RCI analyses typically did not demonstrate improved levels of parental stress or the child's everyday functioning. Parental feedback regarding the study indicated the intervention design was manageable and accessible.

Conclusion: This is the first study to design an IMB informed intervention to improve adherence for paediatric neurological conditions, suggesting future research could continue to apply the model. Targeted recommendation increases indicate that information, motivation, and behaviour skills are all required for sustained and significant increases in adherence. The IMB model components could be incorporated into clinical neuropsychological assessments and used to inform and tailor reports and recommendations. Future studies could consider how the IMB model can be adapted to develop interventions to support improved adherence for different difficulties.

Key words: Paediatric neurological conditions, parents, recommendations, adherence, IMB model, intervention, single-case experimental design, online survey

Introduction

Neurological disorders result from damage to the central nervous system (Child Neurology Foundation, 2019; NHS England), leading to physical and or psychological symptoms and patients require lifelong care. In children such disorders include Sturge-Weber Syndrome (SWS), rare neurocutaneous disorders (e.g. Congenital Melanocytic Nevus, CMN), chromosomal disorders and more common disorders (e.g. epilepsy). Lifelong difficulties can arise in areas such as executive function (MacAllister, Vasserman, Rosenthal, & Sherman, 2014), attention (Dunn, Austin, Harezlak, & Ambrosius, 2003) and memory (Nolan et al., 2004). Parents of children with a neurological condition have to adjust to the diagnosis, their child's condition and on-going health needs. For many, this involves grieving, sorrow (Hobdell, 2004; Hobdell et al., 2007; Yehene et al., 2019) and increased stress due to managing their child's health as well as everyday demands (Cousino & Hazen, 2013; Emerson & Bögels, 2017). Research considering parental stress in the context of their child's condition has typically focused on physical health (Crawford, Garthwaite, & Porter, 2010; Drotar & Bonner, 2009; Klok, Kaptein, & Brand, 2015; Psihogios, Fellmeth, Schwartz, & Barakat, 2019). The complexity of paediatric neurological conditions likely also introduces burden but they have been less researched.

Children with neurological conditions receive care from specialists including paediatric neuropsychologists. Paediatric neuropsychologists complete detailed assessments to inform diagnosis and treatment. Assessments may involve psychometric tests, observations and discussions with those who know the child (Sparrow, 2007). Assessment findings and tailored recommendations are shared in writing with the family and relevant professionals. Recommendations typically use

strengths to support areas of weakness (Klein-Tasman, Phillips, & Kelderman, 2007). Accordingly, the current study defines neuropsychological recommendations as those written by a neuropsychologist referencing behavioural and educational strategies. These will be called recommendations throughout the paper.

Although paediatric neuropsychologists' work can include direct interventions, their recommendations often require support from people with an on-going relationship with the child (Sparrow, 2007). Many recommendations relate to the home, requiring parental support. Given how broad a parent's role is (e.g. attending appointments, organising family life, maintaining a career), having the time to understand recommendations and alter home life can be difficult (Savage, Depompei, Tyler, & Lash, 2005). As such, the style of feedback is important, with research confirming a preference for written feedback (Fallows & Hilsabeck, 2013) that is short, readable with clear results and recommendations (Baum et al., 2018).

Despite the potential benefit of adhering to recommendations (e.g. enhancing a child's experience of and capacity to learn), research indicates adherence rates below 50% (Cheung et al., 2014; Quillen, Crawford, Plummer, Bradley, & Glidden, 2011; Westervelt, Brown, Tremont, Javorsky, & Stern, 2007). Factors related to greater adherence include: parental understanding of the recommendations (Bennett-Levy, Klein-Boonschate, Batchelor, McCarter, & Walton, 1994; Westervelt et al., 2007); recommendations being clear and practical, (Cheung et al., 2014; Westervelt et al., 2007); not overwhelming families with information (Quillen et al., 2011); and relating to patient safety (Westervelt et al., 2007). Paediatric neuropsychologists therefore need to produce understandable reports and include recommendations to benefit the child, but which do not, or are not perceived to, overload the family.

Improving Adherence

Interventions to improve adherence for children with chronic health conditions are often not theoretically driven (Morawska, Calam, & Fraser, 2015), and the National Co-ordinating Centre for NHS Service Delivery and Organisation R and D (NCCSDO) has placed medication adherence on their national agenda (NCCSDO, 2005).

Many theories and models have been developed in relation to behaviour change. For example the Health Belief Model (HBM) suggests adherence relates to a person's desire to be well and their belief an action will improve their illness (Becker, 1974). However, the HBM overlooks access to information or skills. Other models also have theoretical limitations that restrict their utility within this study. Namely, biomedical models of adherence tend to consider patients as passive and behavioural learning models focus more on external skills and the environment (Munro, Lewin, Swart, & Volmink, 2007). One model that overcomes some of these limitations is the Information-Motivation-Behaviour skills (IMB) model (Fisher J. D. & Fisher W. A., 1992). Munro and colleagues (2007) conceptualise this as a cognitive model, whereby an individual's attitudes, beliefs and expectations of future events are important in determining health related behaviour. Although one general limitation of cognitive models is that behavioural components are overlooked, the IMB model places behavioural skills centrally. The IMB model also overcomes the HBM limitation by acknowledging the importance of knowledge. These factors, as well as the model's generalisability and simplicity, led to the IMB model being chosen for the current study.

The IMB model contains three components: 1) information, 2) motivation and 3) behaviour skills, that are tailored to the individual. Information is defined as knowledge specific to the area of interest. Motivation to change incorporates the individual's perception of others' views (e.g. social norms) and their own attitude towards the action. This is understood to be influenced by the perceived behaviour consequence. Finally, behaviour skills relate to the specific skills required to make changes. The authors proposed all three dimensions should be incorporated to effect change; however, the behavioural skills component has been found to mediate the relationship between information and motivation to adherence (Alexander, Hogan, Jordan, DeVellis, & Carpenter, 2017).

One weakness of the IMB model is the absence of a meta-analysis assessing its effectiveness. Further, the model's simplicity could be deemed too parsimonious. However, the generalisability of the model components enable learning from prior research to be incorporated. For example, research has identified that 'information' is required for adherence (e.g. written reports or greater clarification; Fallows & Hilsabeck, 2013; McLoone, Wakefield, Butow, Fleming, & Cohn, 2011; Meth, Calamia, & Tranel, 2016; Quillen et al., 2011), but it is not sufficient, as adult neuropsychological interventions providing information do not improve adherence (Fallows & Hilsabeck, 2013). Further, the motivation component enables consideration of barriers to adherence, which for paediatric conditions includes parental factors (De Civita & Dobkin, 2004). For example, motivation is reduced for recommendations perceived as requiring increased parental effort (Cheung et al., 2014). Finally, interventions can provide the specific behaviour skills to enable adherence in each family's situation without parents having to expend additional effort, thereby overcoming the barrier to adherence noted by Cheung et al. (2014).

A second limitation of the IMB model relates to its limited application beyond adult HIV. However, research is now beginning to apply the model to paediatric populations. Most studies have researched barriers to HIV medication adherence (Dima, Schweitzer, Amico, & Wanless, 2013; Hawkins, Evangelini, Sturgeon, Le Prevost, & Judd, 2016; Rongkavilit et al., 2010) or safe sex behaviours (Kudo, 2013). One study used the IMB model to examine adherence barriers for children with sickle cell disease (Raphael et al., 2013). Only one has developed an intervention to improve adherence for children with HIV, which was effective (Giralt et al., 2019). To date no research has developed an intervention to support adherence for children with neurological conditions.

Research typically assesses specific conditions. However, Morawska et al., (2015) suggest interventions to improve adherence could focus on groups of paediatric conditions due to the similar impact of family context on adherence. Therefore, research findings related to improving adherence for a sample with a rare condition, such as SWS, are likely to be generalisable to other paediatric conditions where neuropsychological recommendations are provided. Considering rare neurological conditions such as SWS is feasible due to the presence of a specialist service within the United Kingdom (UK). This study aims to investigate whether an IMB model intervention could improve the implementation of recommendations within families where a child has SWS.

Current study

This study used SWS as a representative sample for rare paediatric neurological conditions. SWS is a non-inherited neurological condition undetectable until birth, that develops in-utero due to erroneous brain surface blood vessel

development (Sudarsanam & Ardern-Holmes, 2014). Common comorbidities include: epilepsy, intellectual disability and visual field impairments (Sudarsanam et al., 2014). The UK SWS clinic also assesses rare neurocutaneous disorders such as CMN and some chromosomal disorders. CMN results from a genetic mutation and has a high probability of neurological difficulties (Caring Matters Now, 2019). Yearly appointments are offered, where physical, speech and language and neuropsychological needs are assessed. Subsequently, families and relevant professionals receive a detailed report.

An intervention was designed using the IMB model, which aimed to improve the implementation of recommendations through: 1) increasing parental understanding of their child's difficulties and recommendations ('information'), 2) assessing and working with each family's goals by focusing on recommendations pertinent to them ('motivation') and 3) providing clear and specific instructions for each focused recommendation as well as a daily prompt to implement it ('behaviour skills'). To explore the impact of the intervention, specific recommendations were focused on (targeted recommendations). However, to assess whether the intervention would lead to a generalised improvement in adherence, non-targeted recommendations were also assessed.

National guidance states interventions should be tailored to the individual (NICE, 2014), and children included in their care (CQC, 2016). This study therefore sought to develop a tailored intervention, and to include the child. National guidance also recommends gathering information from relevant parties, such as families, when designing intervention services (NICE, 2014). Whilst service development is not the current focus, parental feedback was sought regarding the experience of completing

the intervention, to enable learning for future studies and inform service development.

Aims

1. Investigate whether recommendation implementation among school-aged children with a rare neurological condition can be increased through the application of the IMB model.
 - a. A longer-term aim includes clinical implementation.
2. Investigate whether applying the IMB model leads to changes in:
 - a. Parental stress or burden or
 - b. The child's everyday functioning.
3. Understand families' experience of completing the intervention.

Research Questions

1. Can the implementation of recommendations among school-aged children with a rare neurological condition be improved using an IMB model intervention?
2. Does the intervention lead to improvements in parental stress and the child's everyday functioning?

Hypotheses

1. The frequency of targeted, not non-targeted, recommendations implemented will increase following the intervention.
2. The intervention will lead to positive changes in the child's everyday behaviour.

3. Parental stress will reduce following the intervention.

Method

Design

A single case experimental design (SCED) was used. The independent variable (IV) was a distance-based IMB model intervention. The intervention contained the same components for all families but was tailored to each child. The dependent variable (DV) was the daily number of recommendations each family implemented.

A multiple baseline design (MBD) was followed using a simultaneous replication AB design, where A and B were baseline and intervention phases respectively. Each participant completes both phases of the study, serving as their own control. Replicating the study in multiple participants increased external validity (Onghena & Edington, 2005) and power (Ferron & Sentovich, 2002), and a simultaneous replication design enhanced internal validity (Christ, 2007). Ferron and Sentovich (2002) indicate an MBD with four participants and 20 measurements achieves power of >0.8 . To further enhance internal validity and enable statistical analysis, the baseline duration was randomised across participants (i.e. phase B started on a different day for each participant; Bulté & Onghena, 2008, 2009). Minimum phase lengths were five and fourteen days for phase A and B respectively and the study lasted thirty-one days in total for each participant. Phase B therefore had twelve possible start days (between day six and seventeen). To enable simultaneous design analysis, the final participant began the study before the first participant finished the study (see Appendix A).

To address aim 2, this study used pre (prior to baseline) and post (after the intervention) measures of parental stress and the child's everyday functioning. To address aim 3 brief qualitative information was collected from families about their experience of the study during the debrief.

Participants

All contact with families occurred via telephone to reduce burden on families and maximise recruitment.

Eligibility and Recruitment.

This study aimed to recruit fifteen children receiving outpatient care from the national SWS clinic, aged 5-11years old and a primary caregiver. Table 1 details inclusion and exclusion criteria.

Table 1

Study inclusion and exclusion criteria

Inclusion criteria

Fluent English. Established via telephone conversations and clinic records.

Most recent clinic report provided at least a month and not more than 18months earlier.

Report to contain home-based recommendations.

Child between 5 and 11 years old.

Parent had a mobile phone to receive text message prompts

Exclusion criteria

Children with Full Scale Intelligence Quotient ≤ 70 .

The clinical team identified families for recruitment using child's age, spoken language and time since last report. The researcher completed subsequent recruitment. All data protection and ethical guidelines were adhered to (Appendix B).

Considerable time and effort were required between gaining national ethical approval (October 2017) and local NHS board approval (January 2018). Such delay meant only twenty-one possibly eligible families were screened by the researcher. Eleven families were invited to participate. The researcher was unable to consistently contact six families and one family declined due to time constraints. The remaining four families completed the study. Two children were female and three families nominated the mother and one the father, to be the study respondent. Two children were diagnosed with SWS, one with CMN and one with chromosomal mosaicism 22q11 duplication. Child ages ranged from 6 years 11 months to 11 years 7 months. Demographic information and details to clarify age at diagnosis and co-morbid conditions can be seen in Table 2. Letters, information and forms sent to the families can be found in Appendix C.

Table 2

Participant demographics

Participant	Gender	Age (years)	Diagnosis (age at diagnosis)	FSIQ	Number of appointments a year (approx.)	Co-morbid diagnoses	Number of siblings (additional needs)	Parental education level	Parental working status
1	M	7.6	CMN (birth)	98	7	Attention concerns	4 (1 with ASD)	College	FT
2	F	11.6	SWS (4 years)	83	24	SpLD (reading & writing)	4 (0)	College	Self- employed
3	M	10.5	SWS (birth)	Subscales > 70	6	Possible ADHD	3 (0)	Degree	FT
4	F	6.9	Chromosomal mosaicism 22q11 duplication (6.2 years)	86	24	ADHD	2 (0)	Degree	PT

Key: FSIQ – Full Scale Intelligence Quotient; M – Male; CMN – Congenital Melanocytic Nevus; ASD – Autism Spectrum Disorder; FT – Full Time; F – Female; SWS – Sturge-Weber Syndrome; SpLD – Specific Learning Disorder; ADHD – Attention Deficit Hyperactive Disorder. PT – Part Time.

Instruments

For all measures, higher scores indicate a greater level of difficulty. Where noted some measures enquire about a six-month period. For this study, the pre-measure was informed by the prior six months and the post measure by the study duration.

Characterisation measures

Parenting a child with a chronic health condition is known to bring additional stresses (Cousino & Hazen, 2013) therefore measures of parental and child anxiety and depression were completed with the nominated parent prior to baseline. Results are in Appendix E.

Hospital Anxiety and Depression Scale (HADS)

The HADS (Zigmond & Snaith, 1983) is a 14-item questionnaire. Items score between zero and three, indicating symptom presence or absence from “nearly all the time” to “not at all”. Subscale scores fall within ‘normal’, ‘borderline’ and ‘abnormal’ ranges. Cronbach α is 0.76 and 0.8 for anxiety and depression respectively (Mykletun, Stordal, & Dahl, 2001).

Revised Children’s Anxiety and Depression Scale (RCADS)

The RCADS (Chorpita, Yim, Moffitt, Umemoto, & Francis, 2000) is a 47-item questionnaire and items score between zero (not true at all) and three (very much true). Five anxiety subscales (social phobia, panic disorder, separation and generalised anxiety, and obsessive-compulsive) and a major depression subscale are produced. Cronbach α ranges from 0.61 to 0.83 for each subscale (Chorpita et al., 2000).

Pre and post intervention measures.***Behaviour Rating Inventory of Executive Function (BRIEF) parent report.***

The BRIEF (Gioia, Isquith, Guy, & Kenworthy, 2000) is a measure of executive function for children aged 5-18 years. 72 items are scored 'never', 'sometimes' and 'often', typically pertaining to the last six months. Eight domains are summed along two composites, Behavioral Regulation Index (BRI; inhibition, shifting, emotional control), Metacognitive Index (MI; initiation, working memory, planning, organisation of materials and monitoring) and combined to form a Global Executive Composite (GEC). Cronbach α scores range from 0.82-0.98 (Gioia et al., 2000).

Conners 3rd Edition Parent Short-Form (Conners).

The Conners (Conners, Pitkanen, & Rzepa, 2011) is a measure of children's attention and hyperactivity in relation to the last month. The scale is made up of 43 items, scored between zero (not true at all) and three (very much true). Six subscales are produced: inattention, hyperactivity or impulsivity, learning problems, executive functioning, defiance or aggression and peer relations. Cronbach α ranges from 0.77-0.97.

Parental Stress Scale (PSS).

The PSS (Berry & Jones, 1995) measures parental stress with 18 items scored between one and five (strongly disagree to strongly agree). Parents are instructed to answer the questions in relation to how they feel at that time. Higher scores indicate a higher level of parental stress and are related to lower levels of parental sensitivity, poorer child behaviour and lower parent-child relationship quality (Berry & Jones, 1995). The scale was designed to compare pre and post scores. Cronbach α is 0.83.

Strengths and Difficulties Questionnaire (SDQ).

The SDQ (Goodman, 1999) is a 25 item measure of a child's strengths and difficulties over the previous six months. Items are scored 'not true', 'somewhat true' and 'certainly true'. Four subscales (emotional, conduct, hyperactivity and peer-problems) are summed into a total difficulties score. The remaining subscale relates to prosocial behaviours. Additional items relate to the presence of specific difficulties. If positively endorsed, further questions enquire about distress to the child and area of difficulty. The final question pertains to family burden. Cronbach α is 0.73 (Goodman, 2001).

Pediatric Quality of Life Inventory (PedsQL) Parent report.

The PedsQL (Varni, 1998) measures health-related quality of life. Questions related to the preceding month. The core component covers four domains of functioning: physical, emotional, social and school. For this study the multi-dimensional fatigue questionnaire was also included, which consists of three fatigue domains: general, sleep / rest and cognitive. Each item is scored between zero (never) and four (almost always). Cronbach α for the total PedsQL (not including the fatigue subscale) is 0.9 (Upton et al., 2005).

Daily Outcome Measure

The daily outcome measure was parental response regarding implementation of their individualised recommendations (e.g. "Have you tried to implement recommendation 1 today?"). A concise restating of the recommendation followed (e.g. Laura and Hugh using checklists to help to get ready for school – N.B. anonymised names). Families could choose to answer questions via a paper form

over email or an online survey. All families chose the survey. Parents were prompted to complete the daily outcome measure via a daily text containing a survey link. Responses were 'no', 'partially' or 'yes', scored as one, two and three respectively. This avoided confounding 'no' responses with potential missing data. For each possible recommendation, daily scores ranged from one to three.

The family and researcher chose eight recommendations during the pre-baseline conversation for consideration in the study. This was to balance feasibility and acceptability, whilst providing a meaningful intervention for families. Each recommendation had to be present in the clinic report, relate to a home-based change, and be a recommendation the family wanted to consider.

Intervention

The intervention was based on the IMB model of behaviour change (Fisher, J. D., & Fisher, W. A., 1992) and was developed with families after the baseline phase. Motivation was ensured through families identifying three (of the eight) recommendations to focus on (i.e. targeted recommendations). Typically, families perceived these recommendations as potentially beneficial, but requiring support. Initial conversations with families indicated targeting three recommendations was preferred, to ensure manageability.

Information and behaviour components of the model were provided by discussing the recommendation strategies and relevance in detail. Family context was considered to ensure strategies were appropriate. This led to a co-developed understanding of the recommendations and how the family could implement them.

Targeted recommendations were listed on an A3 poster developed by the researcher as a visual reminder and to ensure involvement of the child. The poster design was informed through the researcher's conversation with each child to determine interests (e.g. favourite colour; see Appendix F).

Following the conversation and poster delivery, the intervention phase started. Families completed the same daily outcome measure as during the baseline. This enabled the impact of the intervention on the three targeted recommendations to be compared to the five non-targeted recommendations.

Procedure

The study procedure is outlined in Figure 1.

Pre-baseline.

Subsequent to parents returning informed consent (and assent) the characterisation (demographics, HADS and RCADS) and pre-study measures (Conners, BRIEF, PSS, SDQ and PedsQL) were completed. The clinic report was reviewed, and eight recommendations were collaboratively identified. Recommendations were tailored to the family and worded concisely. No practical implementation advice was given.

Baseline duration for each family was determined using www.randomiser.org, with six and seventeen as parameters (based on total study duration of thirty-one days and minimum baseline and intervention durations being five and fourteen days respectively).

Baseline.

During the baseline, families completed the daily outcome measure.

Intervention.

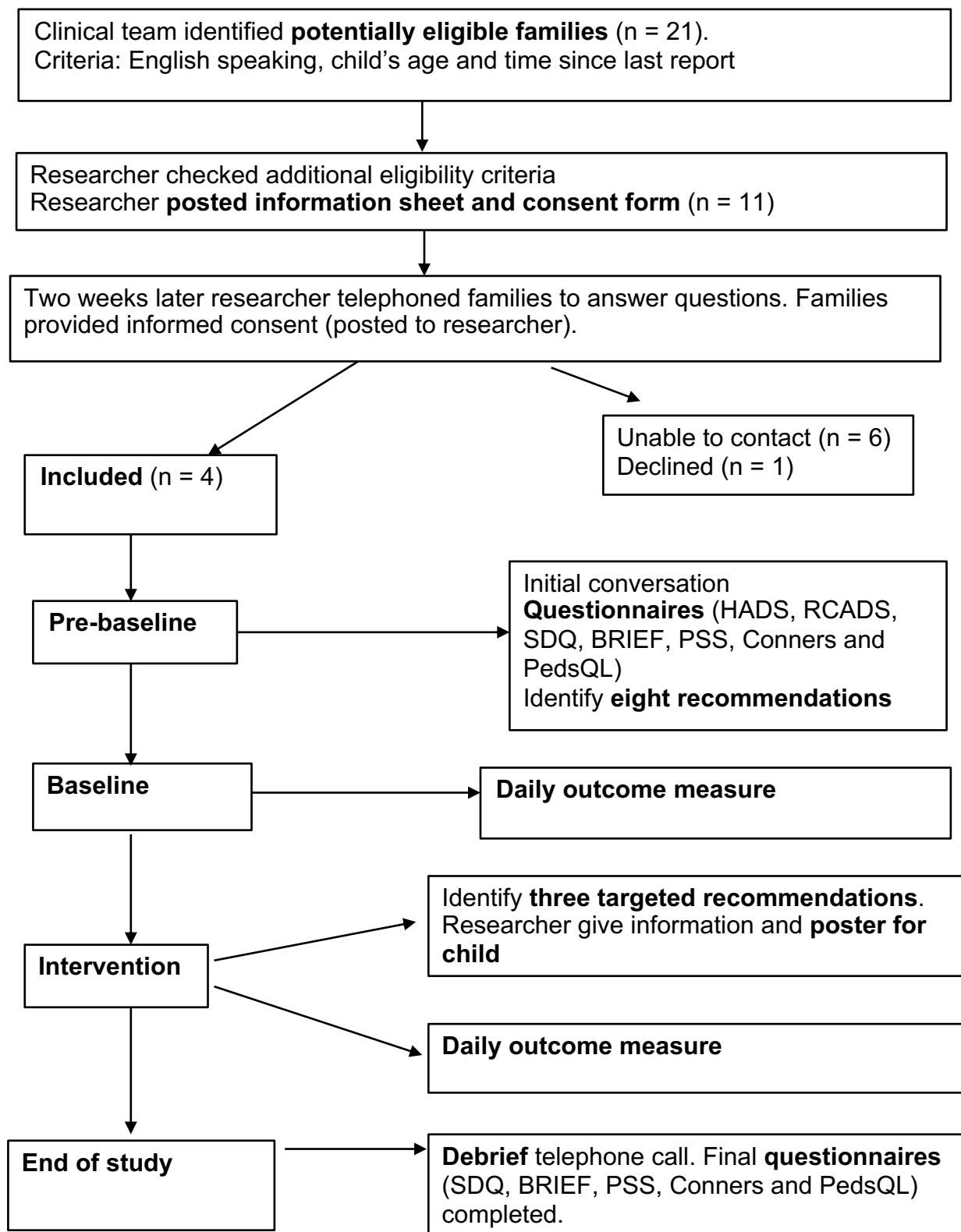
The intervention discussion and poster development were conducted two days prior to the intervention start day to enable delivery of the poster. Families were instructed not to use the poster and new information until the intervention start day. Data collection continued as during the baseline.

For both the baseline and intervention phase, data omissions were monitored by the researcher. Following two consecutive missed days the researcher contacted families to resolve potential difficulties and answer questions.

End of study.

At the end of the intervention families completed final questionnaires (Conners, PSS, SDQ, PedsQL and BRIEF), gave feedback about their experience of the study and received a verbal and written debrief (Appendix C).

Figure 1

Recruitment and study procedure.

Key: HADS – Hospital Anxiety and Depression Scale; RCADS – Revised Children's Anxiety and Depression Scale; SDQ – Strength and Difficulties Questionnaire; BRIEF – Behavior Rating Inventory of Executive Function; PSS – Parental Stress Scale; PedsQL – Pediatric Quality of Life Inventory.

Statistical Methods

Sample characterisation.

Descriptive statistics were used for demographic and characterisation data.

Hypothesis 1: Implementation of targeted recommendations will increase following intervention.

Missing data.

Participant 1 had three missing data points in the baseline phase. Participants 3 and 4 had twelve and nine missing data points respectively, split across both phases. Crucially neither exceeded the recommended 50% of total data (Onghena, 2019). Forgetting was the dominant reason provided. Missing data was imputed with the broadened phase median, which enabled an improved estimate of central tendency (Morley, 2018) and aspects of visual analysis (VA; e.g. calculation of trend).

Data cleaning.

Initial visual inspection of the data showed variability and a non-linear trend for most participants (see Appendix G). Following guidelines by Morley (2018), data was smoothed by calculating a running median of three successive points (RM3).

Analysis.

Frequency of recommendation implementation was analysed using VA and a randomisation test (RT). Analyses were completed separately for targeted and non-targeted recommendations.

VA is a necessary and helpful stage in analysis (Lundervold & Belwood, 2000; Morley, 2018). VA followed guidelines by Morley (2018) and was completed within Excel™. The split-middle method was used to explore trend as it is not influenced

by possible outliers. The broadened median was calculated as the measure of central tendency as it uses more data points compared to the standard median. The overall range was used to assess variability as data points were not spread across a large range (e.g. mostly within one to two points). Graphs to visualise overlap of data between phases were produced as this uses all data, rather than focusing solely on the point of change. Graphs displaying the median, range and overlap of data between phases can be found in Appendix G.

The daily outcome measure was scored on a small scale (e.g. one to three) therefore, responses for all targeted and non-targeted recommendations respectively were summed for each day. Scores per person per day could range from three to nine for targeted recommendations and five to fifteen for non-targeted recommendations. Pertinent to VA, the axes are different across graphs for targeted and non-targeted recommendations preventing a direct comparison. However, the pattern of responses was visually compared to assess the specificity of the intervention's effect. This addresses hypothesis 1 as the intervention was hypothesised to only improve targeted recommendation implementation.

The estimated frequency of pre-baseline recommendation implementation was scored retrospectively using the initial conversations between researcher and family and added to VA graphs. This provided an indication of the impact of identifying recommendations at the start of baseline.

The intervention effect on recommendation implementation was investigated using a RT comparing the difference in mean frequency of recommendation implementation pre and post intervention. RTs are not based on assumptions of

homogeneity (Bulté & Onghena, 2008, 2009; Edington & Onghena, 2007) and determine the probability of the observed data occurring given all possible data. The test statistic is computed within each possible randomisation distribution, where the computation is not overly demanding (Heyvaert et al., 2017; Morley, 2018). The current study had nearly 500,000 possible randomisation distributions. Therefore, a Monte Carlo simulation with 1,000 randomisation distributions was computed (Bulté & Onghena, 2008, 2009; Heyvaert et al., 2017; Morley, 2018).

While the test statistic determines whether the null hypothesis can be rejected, effect sizes quantify the intervention's effectiveness and should be cited more frequently in SCEDs (Crawford et al., 2010). The Non-overlap of All Pairs (NAP; Parker & Vannest, 2009) was used in this study as it uses all available data.

The alpha level for the RT was 0.05 in line with traditional psychological research. However, an alpha value of 0.08 was considered suggestive of a significant result due to it being the lowest *p*-value obtainable for this study based on the twelve possible phase change days (i.e. $1/12 = 0.08$; Morley, 2018).

All analyses were completed using R software from the Comprehensive R Archive Network website (CRAN; www.cran.r-project.org) and functions written by Bulté and Onghena (2008, 2009, 2013). Calculations were computed on raw data. The randomisation and effect size test were calculated for each participant and participants as a group. This enabled analysis of change per person, in line with the goal of SCEDs, whilst combining participants increased the power to detect an effect.

Hypothesis 2 & 3: The intervention will lead to positive changes in the child's behaviour and parental stress.

Hypotheses 2 and 3 were addressed using the Reliable Change Index (RCI; Morley & Dowzer, 2014). This is accessible and useful for intervention outcomes in SCEDs (Busse, McGill, & Kennedy, 2015).

For hypothesis 2 the RCI analysed a pre to post change in the child's functioning on the BRIEF, SDQ, Conners and PedsQL. For hypothesis 3, PSS pre and post scores were analysed. The SDQ question pertaining to family burden cannot be analysed using the RCI; however raw scores were compared to assess change.

Qualitative information

Data collected during the debrief with families was analysed using content analysis (CA; Byrman, 2012). The brevity of information prohibited more detailed analysis; however, CA enabled pertinent themes to be established and their frequency counted. Conversations were not recorded due to ethical constraints; therefore, themes were established from debrief notes.

Results

Four families completed all aspects of the study. Demographic and characterisation profiles can be found in Table 2 and Appendix D.

Hypothesis 1

Targeted recommendation results will be considered prior to non-targeted recommendations and the two conditions subsequently compared.

Visual Analysis.***Targeted recommendations.***

Figure 2 displays trend targeted recommendation implementation for each participant.

Participant 1's data shows a gradual decline throughout the baseline following an initial increase from pre-baseline. Following the intervention, implementation increased to ceiling, which was generally sustained.

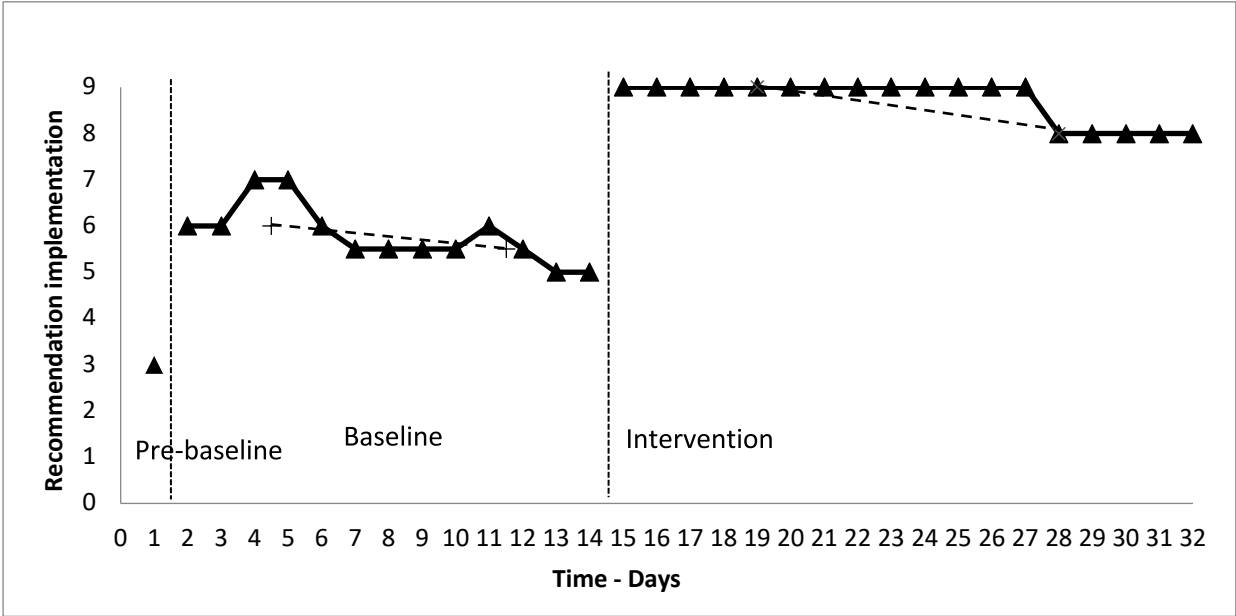
Participant 2's data had clear implementation increases at baseline and intervention. The baseline increase was sustained across the phase (although baseline was notably shorter than other participants). The intervention increase took implementation to ceiling, which was sustained for approximately two weeks, following which, more variability was present.

Participant 3 showed a clear increase at the beginning of baseline; however, implementation quickly declined to lower than pre-baseline. Despite an overall declining trend across baseline, implementation slightly increases halfway through. The intervention phase demonstrates a further increase, with implementation remaining above trend baseline level.

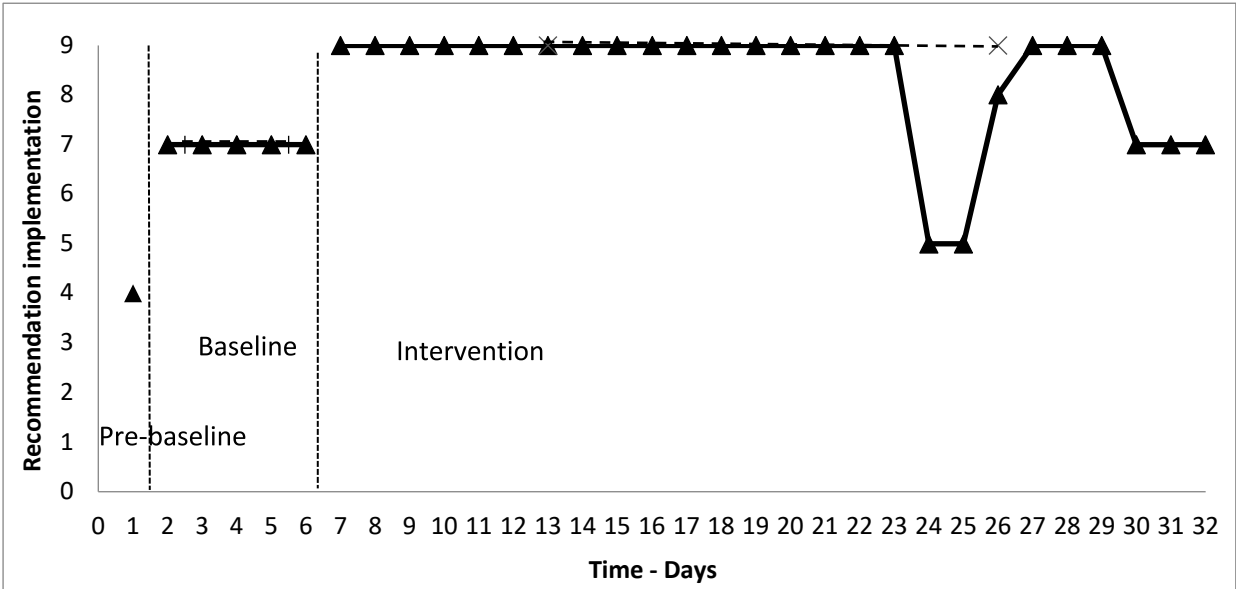
For participant 4, implementation remained stable across the baseline phase, in line with pre-baseline rate. However, there was a notable increase from baseline to intervention, with variation in daily implementation.

Figure 2

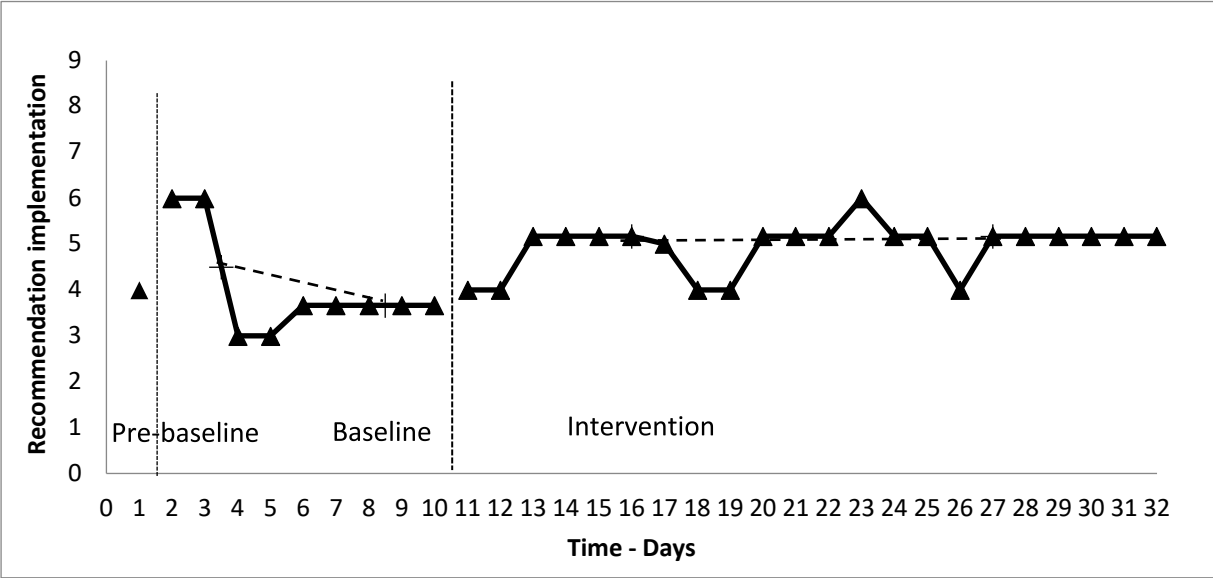
Trend targeted recommendation implementation for all participants



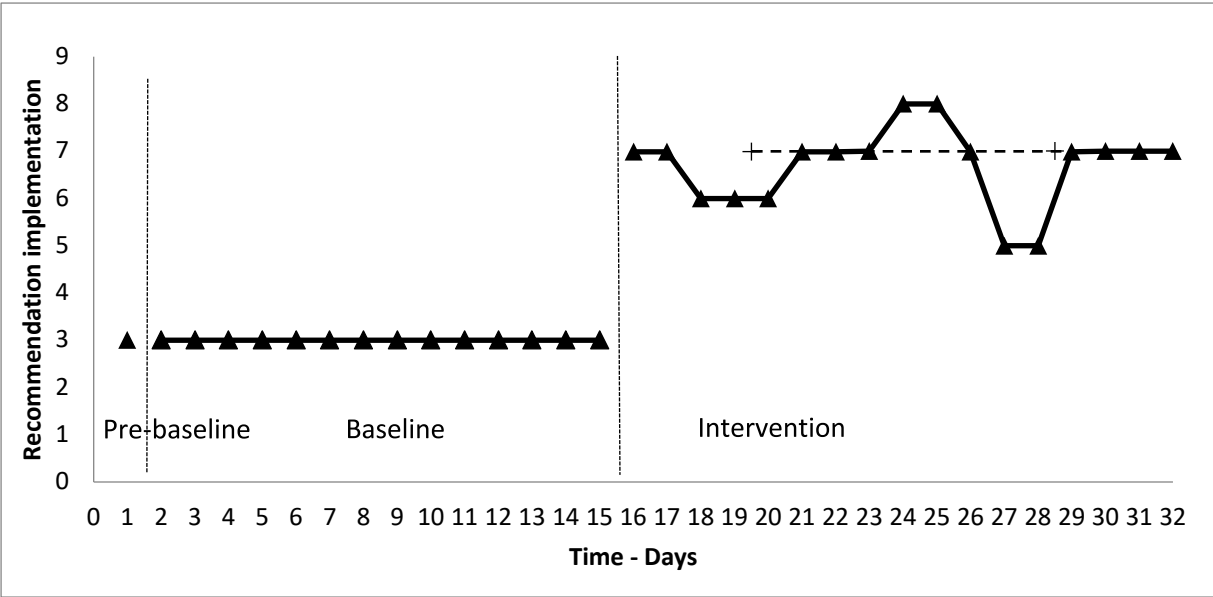
Participant 1: Trend targeted recommendation implementation



Participant 2: Trend targeted recommendation implementation



Participant 3: Trend targeted recommendation implementation



Participant 4: Trend targeted recommendation implementation

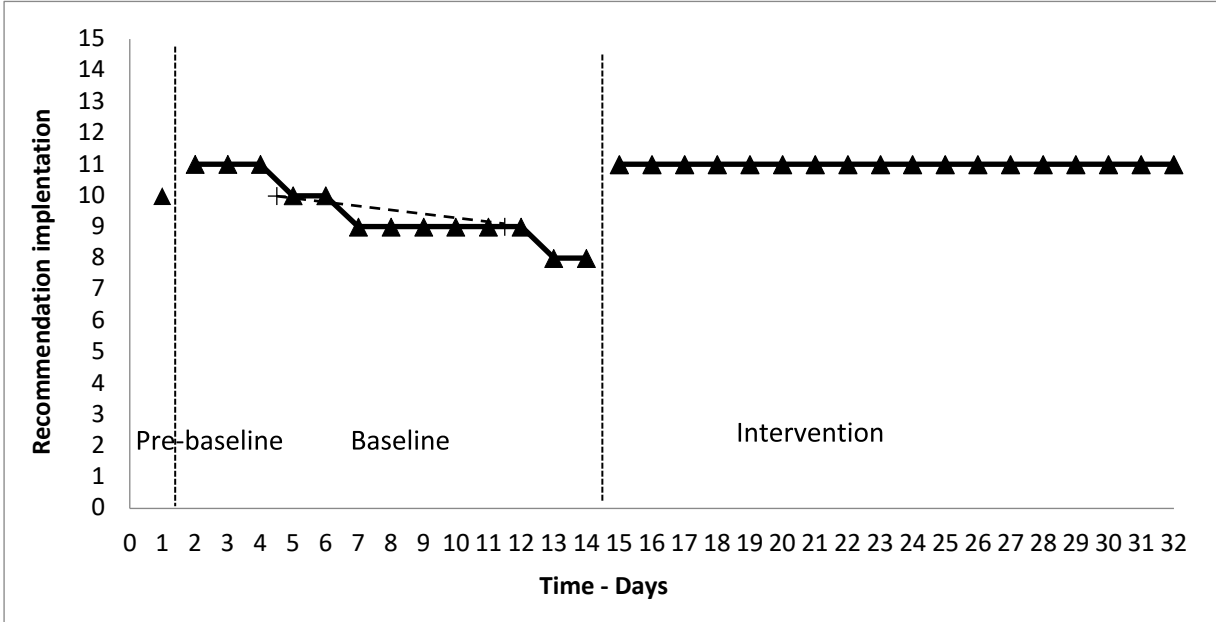
Non-targeted recommendations.

Figure 3 displays trend non-targeted recommendation implementation for participants. For participants 1, 2 and 3 there was an increase in recommendation implementation from pre-baseline to baseline. This increase was maintained for participant 2, where scores remained at ceiling throughout the study. However,

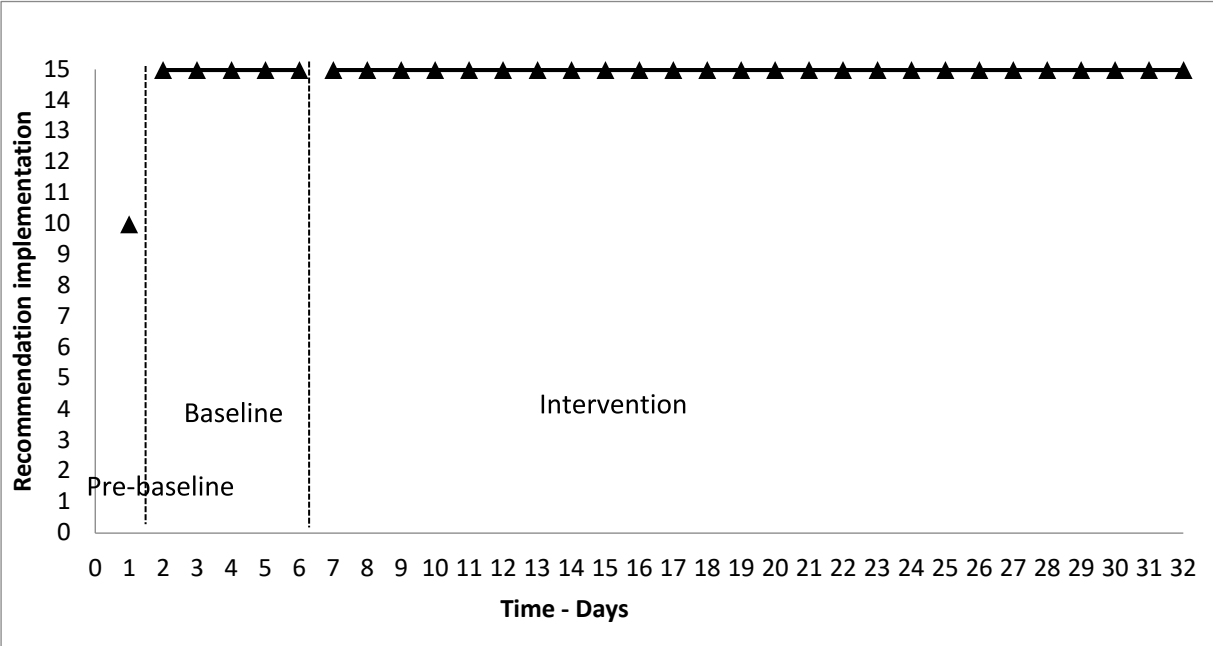
participants 1 and 3 showed a gradual decline in implementation during the baseline phase to below pre-baseline level. Participant 1’s implementation increased and was sustained, at the intervention phase change. Participant 3’s implementation increased gradually across the intervention phase. Participant 4 had a notable implementation decline between the pre-baseline and baseline phases. Although there was a small increase in implementation across the baseline, implementation remained below pre-baseline level. At intervention onset, there was a stepwise increase in implementation to pre-baseline level, which was maintained to study completion.

Figure 3

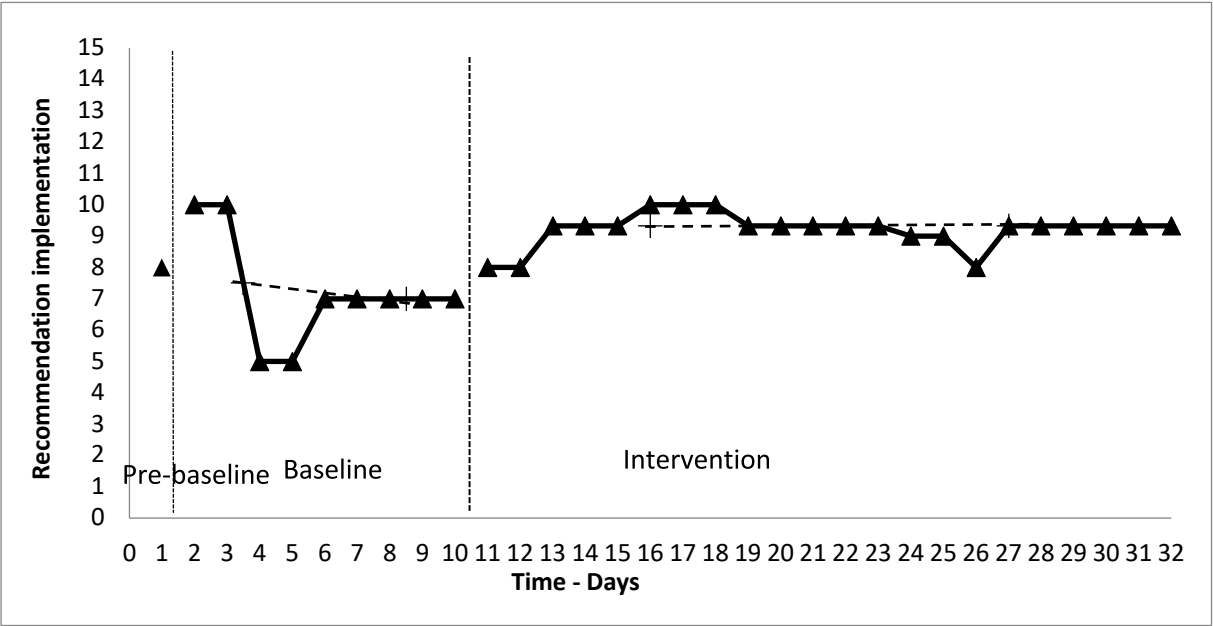
Trend non-targeted recommendation implementation for all participants.



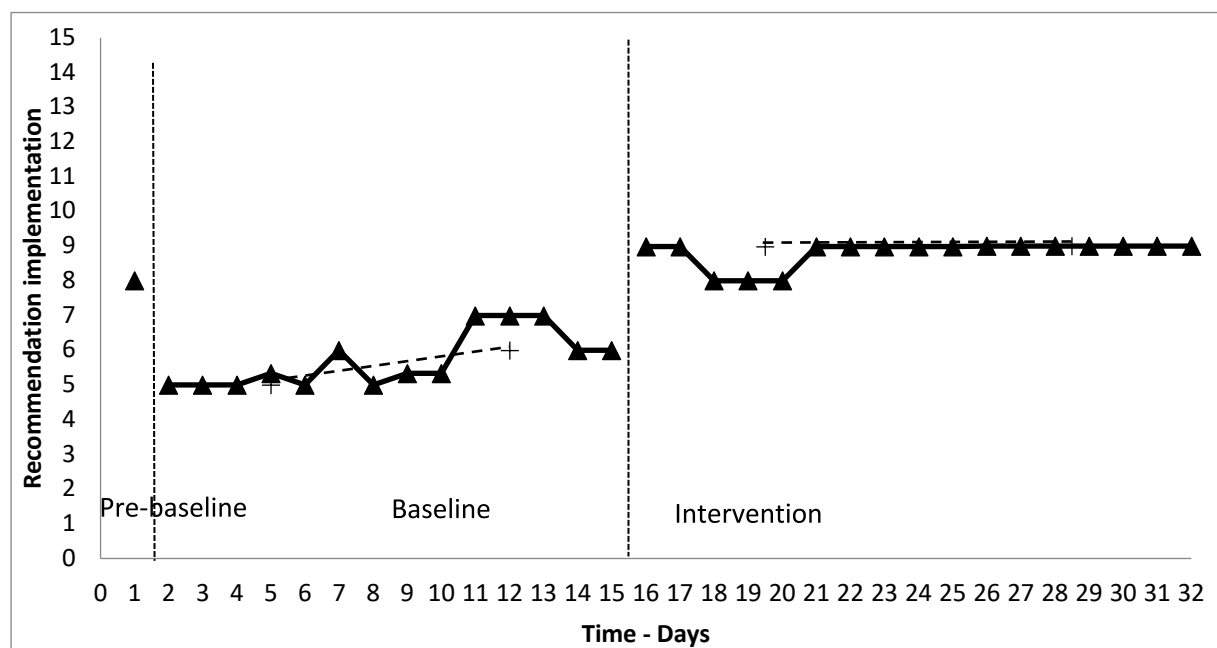
Participant 1: Trend non-targeted recommendation implementation



Participant 2: Trend non-targeted recommendation implementation



Participant 3: Trend non-targeted recommendation implementation



Participant 4: Trend non-targeted recommendation implementation

Intervention specificity.

Participant 1 demonstrated a 33.3% increase from pre-baseline to baseline and from baseline to intervention for targeted recommendations. In comparison, non-targeted recommendation implementation gradually declined during baseline. Although implementation did increase following the intervention, this was a smaller proportional change than for targeted recommendations (20%).

Implementation of targeted recommendations for Participant 2 went from partial to full after the intervention, representing a 22.2% increase. In contrast, non-targeted recommendations were at full implementation from the baseline phase throughout the study.

Participant 3 had a similar pattern for both targeted and non-targeted recommendations. Implementation increased from virtually none during baseline, to partial throughout the intervention.

Participant 4 showed a 44% increase in targeted recommendation implementation between baseline and intervention, reflecting a change from no implementation during baseline, to partial during the intervention. A similar but less distinct change was present for non-targeted recommendations (e.g. 20% increase).

Randomisation and effect size test.

Table 3 displays the mean frequency, NAP and RT of each participant for targeted and non-targeted recommendations.

Targeted recommendations.

NAP findings showed individual participants and the group of participants increased implementation, as hypothesised. Most effect sizes were medium or large (>0.66), suggesting at least a moderate difference between baseline and intervention phase data.

RTs for each participant did not show statistically significant changes between the frequency of recommendation implementation in the baseline compared to the intervention phase. However, a significant result was observed for the group ($p=0.008$). This appears driven by results from participants 1 and 2 as p -values were notably lower compared to participants 3 and 4. Notably, although the VA for participant 4's targeted recommendations indicates a large difference between phases, this participant had nine missing data points. Although imputed for the VA, the RT is calculated using raw data, meaning these data points were lost.

Overall, the results suggest a significant increase in implementation rates of targeted recommendations between baseline and intervention phases for the group. This demonstrates partial support for hypothesis 1.

Non-targeted recommendations.

NAP results for participants 1, 2, and 4 and the group combined, showed an effect size suggestive of an increase in non-targeted recommendation implementation between baseline and intervention phases. Participant 3's NAP results were just below cut-off. Results for participants 2 and 3 showed a 'weak' effect, participant 1 and the group overall a 'medium' effect and participant 4, a 'large' effect. However, no significant RT results were found, indicating no change in implementation rates between baseline and intervention phases.

Table 3

Frequency of recommendation implementation, effect size and randomisation test

Participant	Phase duration (days)		Mean recommendation frequency (SD)		NAP	P-value	Statistically significant?
	Baseline	Intervention	Baseline	Intervention			
Targeted recommendations (possible score range is 3-9)							
1	13	18	5.50 (1.02)	8.67 (0.59)	0.994	0.078	No*
2	5	26	7.8 (1.1)	8.42 (1.2)	0.673	0.084	No*
3	9	12	4.37 (1.7)	4.8 (1.1)	0.518	0.676	No
4	15	16	2.99 (0.001)	6.47 (1.12)	1	0.239	No
Combined	42	72	4.68 (1.84)	7.12 (1.91)	0.796	0.008	Yes

Non-targeted recommendations (possible score range is 5-15)							
1	13	18	9.15 (1.28)	10.94 (0.24)	0.886	0.081	No*
2	5	26	15 (0)	15 (0)	0.5	1	No
3	9	12	8.11 (2.98)	8.83 (2.15)	0.488	0.589	No
4	15	16	5.87 (1)	8.82 (1.07)	0.942	0.532	No
Combined	42	72	8.51 (3.24)	11.22 (2.95)	0.704	0.187	No

Key: SD – Standard Deviation; NAP – Non-overlap of All Pairs. Tentative NAP ranges: 0-0.65 weak effect; 0.66-0.92 medium effect; 0.93-1.00 large effect (Parker & Vannest, 2009).

* results suggestive of significant result using alpha level of 0.08 (lowest obtainable *p*-value for current study due to the 12-day phase change).

Hypothesis Two and Three

Table 4 displays pre and post study questionnaire scores and RCI results (Appendix G contains subscale scores).

Hypothesis two predicted the intervention would lead to positive child behaviour changes. PedsQL, Conners and SDQ RCI results do not indicate any change in the child's behaviour. RCI for the BRIEF also did not show a significant change for participants 2, 3 or 4. However, the BRIEF BRI for participant 1 showed a significant improvement.

Hypothesis three predicted parental stress would reduce following the intervention. PSS RCI results for participants 1, 2 and 3 do not support this. However, participant 4 (who had the highest pre-PSS score) showed a significant

improvement, suggesting their parental stress reduced during the study. In addition, the SDQ burden question was examined. Answers for participants 1, 2 and 4 did not change during the study. However, participant 3 reduced their sense of burden from 'quite a lot' to 'only a little' across the study.

These findings suggest the intervention generally did not lead to improvements for the child's behaviour or functioning or reduced parental stress. However, the results do indicate the intervention could lead to clinically significant improvements for some parents and children.

Table 4

Questionnaire scores and RCI findings

	Participant 1			Participant 2			Participant 3			Participant 4		
Questionnaire	Pre	Post	RCI	Pre	Post	RCI	Pre	Post	RCI	Pre	Post	RCI
	change?			change?			change?			change?		
PedsQL	86	80	No	67	61	No	51	38	No	34	30	No
BRIEF: BRI	82	76	Yes	71	71	No	69	68	No	67	72	No
BRIEF: MI	71	67	No	73	69	No	64	57	No	87	83	No
BRIEF: GEC	71	72	No	74	71	No	67	62	No	81	81	No
Conners	42	43	No	30	32	No	41	37	No	52	51	No
SDQ	25	24	No	12	16	No	20	14	No	15	15	No
PSS	40	41	No	39	40	No	44	38	No	51	39	Yes
SDQ burden	2	2	NA	1	1	NA	2	1	NA	2	2	NA

Key: BRIEF – Behavior Rating Inventory of Executive Function; BRI – Behavioral Regulation Index; MI – Metacognition Index; GEC – Global Executive Composite; PedsQL – Pediatric Quality of Life Scale; PSS – Parental Stress Scale; RCI – Reliable Change Index; SDQ – Strengths and Difficulties Questionnaire. N.B. Lower scores indicate

improvements. SDQ burden score – 0 = Not at all, 1 = Only a little, 2 = Quite a lot, 3 = A great deal.

Qualitative Findings

Table 5 lists the extracted themes and frequency of report. Most findings were positive, with parents reporting improved confidence. Posters were enjoyed by the children and three families intended to continue displaying it. Further, all families intended to maintain implementing recommendations from the study.

All families found the daily outcome measure manageable and accessible. However, parents stated their responses felt repetitive and one parent indicated they may not have been the most appropriate respondent. One parent reported the questionnaires were not capturing their child's progress.

Finally, all families reported the study increased focus and that discussions regarding the clinic report were essential to enable change because they improved understanding. In addition, one family suggested the readability of reports could be improved.

Table 5

Parental feedback themes

Theme	Frequency
Positive	
The survey was manageable timewise, accessible and quick.	4
Children liked the posters, feeling proud and motivated.	3
Posters helped focus the family.	1
Poster to stay up after the study.	3

Theme	Frequency
Intervention helped parent to focus and served as a reminder for recommendations.	3
Made whole family aware of recommendations, enabled a conversation and shared the responsibility.	2
Felt able to continue to work on some recommendations.	4
Discussion was important element of intervention as increased understanding of child's needs and how to establish a new routine.	3
Improved parent's confidence (e.g. how to manage changes to routine).	2
Increased parent's pride and reduced overwhelmed feelings.	1
Negative	
Questions felt repetitive after a few days.	2
Desired clarity about when study end date.	1
Questionnaires not capture change family noticed in their child.	1

Discussion

This study investigated the effect of a distance based, IMB informed intervention on recommendation implementation for families whose child has a rare neurological condition. Hypothesis one predicted increased targeted recommendation implementation following the intervention. Group RT supported this through a statistically significant implementation increase of targeted, not non-targeted, recommendations. VA demonstrated further support with a clearer pattern and greater increases in recommendation implementation for targeted, compared to

non-targeted, recommendations. Partial increases in non-targeted recommendation implementation, as with participant 2, may represent a generalisation of knowledge from targeted recommendations or that implementation of non-targeted recommendations was more straightforward.

Hypothesis two predicted improved child behaviour following the intervention and overall was not supported. The relatively short nature of this study may have prevented changes being detected within questionnaires. However, the results could reflect that the specific behaviours focused on in recommendations were not included in the questionnaires, as these enquired about behaviour domains. However, participant 1 had a significantly improved BRIEF BRI score. Notably, the behaviours that improved for this child were specifically assessed on the BRIEF (e.g. inhibiting extreme emotions) and could have been related to the targeted recommendation strategies. Therefore, the improved BRI could reflect effective strategy use.

The final hypothesis predicted reduced parental stress following the intervention. Although generally unsupported by RCI analysis, participant 4 showed significantly reduced parental stress. This parent's pre-PSS score was the highest in the study with reports of exhaustion. Successful stress reduction may therefore have been more feasible. Further, their child was diagnosed less than a year before the study, suggesting this parent may have been experiencing heightened stress while adjusting to their child's diagnosis and health care needs (Cousino & Hazen, 2013; Emerson & Bögels, 2017). Whilst this indicates interventions for parents experiencing parental stress may be most effective soon after diagnosis, this timing could increase stress for some parents through the addition of another, well

intentioned, demand. Therefore, intervention content and timing must be tailored to the family. Finally, two families' PSS scores increased by one point across the study. This could suggest that focusing on their child's needs and making necessary changes increased parental stress to a small degree. However, it is important to note that the RCI findings did not indicate a statistically significant increase and therefore conclusions here should be tentative.

Theoretical Implications

Previous IMB model applications focused on HIV related behaviour (Dima et al., 2013; Hawkins et al., 2016; Rongkavilit et al., 2010). Giralt et al. (2019) recently demonstrated the model's efficacy in improving adherence to a new medication among children with HIV. The current study represents the first to design an IMB informed adherence intervention for children with a rare neurological condition. This model was appropriate and applicable to this population, with families expressing increased awareness, insight and confidence. Further, this study found significant and sustained increases in implementation for targeted, but not non-targeted, recommendations, supporting the need for all three IMB model components to significantly improve adherence (Fisher J. D. et al., 1992). The current study therefore provides support to the IMB model's suitability for designing adherence interventions (Munro et al., 2007), and evidence for its applicability beyond medication and HIV.

Despite these strengths, the appropriateness of applying an intervention for families where a child has a rare neurological condition needs consideration. For example, the IMB model requires individuals (in this study, parents) to be able to

consider the information already provided, their motivation and develop behavioural skills. However, for some families where a child has a neurodisability, this may not be feasible due to already strained emotional and practical resources. Therefore, any intervention may be secondary to seeking practical family support (e.g. appropriate housing). Additionally, amendments to an IMB intervention may be required such as adapting information to facilitate understanding and motivation, and underpinning behaviour change through access to local resources (e.g. support groups). These adaptations would need to consider family need, local provision, clinical limitations and research aims. Notably these adaptations highlight the importance of considering these factors when applying theoretical models to clinical practice.

This study was carefully designed to avoid dissemination of intervention information pre-baseline. However, the increases in implementation of non-targeted recommendations as well as the increases for both recommendation types from pre-baseline to baseline, do suggest that simple information (i.e. reviewing the clinic report and the recommendations) and enhancing motivation (i.e. focusing on fewer recommendations) can benefit some families. This supports the need for information to increase parental understanding of the recommendations and reduce parental load (Bennett-Levy et al., 1994; Fallows & Hilsabeck, 2013; McLoone et al., 2011; Meth et al., 2016; Quillen et al., 2011; Westervelt et al., 2007) to improve adherence. However, the significant improvements seen only for targeted recommendations involved all components of the model through: i) increasing parental understanding with detailed conversations considering the family context, ii) increasing motivation through focusing on specific recommendations and iii) the addition of behaviour skills relevant to the family context and presented in a visual reminder. This further

demonstrates support for the IMB model. Prior research had identified the behaviour skills component as particularly crucial, as it mediates the relationship between information and motivation (Alexander et al., 2017); however, this was not possible to analyse in the current study.

Parental feedback regarding the study provided further support for using the IMB model with this population. Verbal discussions were reported as beneficial by providing parents time to consider and ask questions regarding the recommendations. Parents also reported how the children enjoyed the posters, indicating the benefit of including the children, in line with national guidance (CQC, 2016).

Study Limitations

One study limitation relates to the breadth of recommendations included. Initial conversations with families noted numerous relevant recommendations within the report. To ensure motivation (key to the intervention), eight recommendations related to any neuropsychological domain, were chosen. For example, for one family sleep was relevant, whereas other recommendations related to reward charts or communication style. This breadth could have impacted the detectability of behaviour change in the questionnaires. Future studies could choose a more specific measure of behaviour or functioning depending on the recommendations, which was not possible here due to ethical constraints. Alternatively, studies could seek to recruit children with similar neuropsychological difficulties.

Additionally, ideally the allocation of recommendations as targeted or non-targeted would have been randomised and contained an equal number of recommendations in each. However, this was not practical as the study was focused

on supporting families through prioritising their goals. Three targeted recommendations were chosen through conversations with families and clinicians to balance motivation and manageability. Indeed, parental feedback stated the intervention was accessible and manageable, supporting this choice.

Another limitation was the study duration of thirty-one days, which was chosen to ensure study accessibility whilst providing time for families to make changes. However, this is a short timeframe (a short-term intervention is defined as < 6 weeks; NICE, 2014) for changes to result in improvements in a child's everyday functioning. This study could therefore have been improved with a follow-up to repeat questionnaires (e.g. three months). Time constraints prevented this within the current project.

Finally, to reduce burden on families, this study did not complete the pre and post questionnaires between baseline and intervention. The current RCI findings do not indicate the inclusion of measures at this timepoint would have shown significant findings. However, future studies using a measure of behaviour or functioning specific to the recommendation domain, could add this timepoint. This would enable analysis regarding potential change during baseline or after the intervention.

Study Strengths and Future Research

SCED methodology is particularly applicable in clinical settings and for rare disorders (Morley, 2018), and its implementation enabled a powered and appropriate study. This study adds to the growing body of research utilising SCED (Morley, 2018) and the investigation of a group of rare conditions was justified (Morawska et al., 2015). Future studies may continue to benefit from employing SCED methodology transdiagnostically. However, increasing the sample size for

each disorder would increase power through replication and enable disorder specific conclusions. The heterogeneity present in this study reflects the inherent intricacies and challenges of clinical recruitment.

Use of a distance-based intervention was an additional strength of this study, being practical for families and clinicians. The study used readily available means (e.g. phones and post), to reduce exclusion rates. Study results indicate this style of intervention enhanced recommendation implementation in an acceptable way for families. Implementing this within clinics and future studies could be a cost and time effective method to maximize patient benefit following neuropsychological assessments. Consequently, it could reduce repeated clinic visits and increase clinic efficiency.

Another strength was the first use of an IMB informed intervention to improve adherence for children with neurological conditions. Previous use of the model to develop an intervention for children has focused on HIV (Giralt et al., 2019). Future research to improve adherence, regardless of disorder, could therefore use this model as a theoretical underpinning, as well as a practical tool in intervention design.

A final study strength was the inclusion of parental stress, which is understood to be related to adherence, but the specific relationship to adherence is less well researched (see Literature Review). Future studies could advance understanding regarding whether adherence interventions also impact parental stress through a longer follow-up of parental stress or using different parental stress measures (e.g. Pediatric Inventory for Parents, PIP; Streisand, Braniecki, Tercyal & Kazak, 2001) .

Clinical Implications

Findings from this study suggest that whilst clinicians are able to appropriately assess children and provide recommendations, this could be improved with discussions to establish the family context, main areas of difficulties and current skills employed by the families. The inclusion of children in these conversations is recommended (CQC, 2016). Clinicians could subsequently incorporate this knowledge into their reports and seek verbal feedback with families to ensure all aspects of the IMB model are included. Ensuring families feel able to, and are supported to, implement recommendations is important as recommendations from paediatric neuropsychologists often require on-going involvement from families (e.g. establishing a routine). Such an intervention, a simple but focused change in service delivery, could improve adherence as the collaboration ensures recommendations are relevant and meaningful and reduces parental effort to 'translate' them into their lives.

In line with this and parental feedback in this study, services could improve the readability and utility of their reports. Guidelines for psychologists emphasise the importance of using clear language (American Psychological Association, 1992, 2002) and whilst experts suggest brief reports (Donders, 1999) there are no clear guidelines regarding structure or organisation. However, a recent paediatric neuropsychology service trial demonstrated reports can be more accessible to families and other professionals and less costly to services (e.g. fewer writing hours; Baum et al., 2018).

A final improvement is for services to ensure psychoeducation regarding the child's disorder and specific difficulties is provided to families. This should account

for the family context and the specific skills required to implement strategies. Such improvements are likely to require improved integration and communication between community and hospital based out-patient care within NHS trusts.

Conclusion

The current study provides support for designing interventions to improve neuropsychological adherence based on the IMB model. Detailed discussions focusing on the relevance and meaning of recommendations within the family's context, as well as how to implement them, enabled families to embed the recommended strategies. Future studies should continue to research the applicability of the IMB model for children presenting with particular neuropsychological difficulties.

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Appendix A: Methodology design and analysis

Table 6

Steps to design and analyse SCED (Bulté & Onghena, 2008, 2009)

Steps to design and analyse a single case MBD experiment	Decisions for current study
<i>Decide study design</i>	AB phase design across subjects, with simultaneous replication.
<i>State null and alternative hypothesis</i>	<i>Null hypothesis:</i> Number of recommendations implemented, and parental stress will not change following the intervention.
<i>Choose appropriate test statistic</i>	<i>Alternative hypothesis:</i> Following the intervention, the frequency of recommendations implemented will increase and parental stress will reduce. <i>Test statistic:</i> RT to assess difference in mean frequency of recommendation implementation between Phase A and B.
<i>Determine level of significance and number of measurements</i>	Alpha 0.05 will be used in line with traditional psychological research. The total number of measurements (across both phases) will be 31, with a minimum of five data points for Phase A and 14 for Phase B, leading to 12

	possible phase change days for each participant.
<i>Randomisation schedule</i>	The randomisation schedule (start points for the intervention) was determined prior to the study using www.randomiser.org .
<i>Data collection and calculation of observed test statistic</i>	Data was analysed in R software using functions specifically for MBD studies (Bulté & Onghena, 2008, 2009).
<i>Constructing the randomisation distribution</i>	RTs require all permutations of the data to be considered. However, this is computationally demanding and therefore a “Monte Carlo” simulation was used with 1000 possible permutations.
<i>Assessing the p value</i>	Calculating ‘exact’ p value is too demanding due to the number of possible permutations. Therefore, the p value was calculated using the test statistic within the randomisation distribution.

Key: RT – randomisation test

Appendix B: Ethical approval (National, Local and Exeter University)

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

09 October 2017

Dr Jessica Watts
1 Webbers
Bishops Lydeard
Taunton
TA43QX

Dear Dr Watts

Study title:	An investigation into the impact of a short-term, distance based intervention on the implementation of neuropsychological recommendations for families with a child who has a complex physical health condition.
REC reference:	17/LO/1185
Protocol number:	NA
IRAS project ID:	220848

Thank you for your letter of 30 September 2017, responding to the Proportionate Review Sub-Committee's request for changes to the documentation for the above study.

The revised documentation has been reviewed and approved by the sub-committee.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA Approval (England)/ NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will

be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" above).

Approved documents

The documents reviewed and approved by the Committee are:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Interview schedules or topic guides for participants [Guide for initial conversations with families]	1	21 June 2017
IRAS Application Form [IRAS_Form_29062017]		29 June 2017
IRAS Checklist XML [Checklist_17092017]		17 September 2017
Letter from sponsor [Sponsor confirmation letter]	1	19 June 2017
Letters of invitation to participant [Invitation letter]	1	21 June 2017
Letters of invitation to participant [Invitation letter_to use]	2	24 September 2017
Other [Schedule of events]	1	21 June 2017
Other [Statement of activities]	1	21 June 2017
Other [Debrief sheet]	1	21 June 2017
Other [SDQ questionnaire]	1	04 April 2017
Other [RCADS questionnaire]	1	04 April 2017
Other [PSS questionnaire]	1	04 April 2017
Other [PedsQL]	1	04 April 2017
Other [Example response sheet]	1	22 June 2017
Other [Sarah Aylett CV]	1	22 June 2017
Other [Child PIS]	1	21 June 2017
Other [Sponsor letter to Chief Investigator]	1	19 June 2017
Other [Professional Indemnity]	1	19 June 2017
Other [Public Liability]	1	19 June 2017
Other [Child assent form]	1	21 June 2017
Other [PIS_v2_track changes]	2	24 September 2017
Other [Invitation letter_track changes]	2	24 September 2017
Other [Research protocol v2_track changes]	2	24 September 2017
Participant consent form [Consent form]	1	21 June 2017
Participant information sheet (PIS) [PIS]	2	24 July 2017
Participant information sheet (PIS) [PIS_to use]	2	24 September 2017
Referee's report or other scientific critique report [Project proposal]	1	23 June 2017

(for thesis) reviewed by university]		
Research protocol or project proposal [Research protocol]	1	23 June 2017
Research protocol or project proposal [Research protocol_to use]	2	24 September 2017
Sample diary card/patient card [Example Visual Aid]	1	27 June 2017
Sample diary card/patient card [Example Visual Aid]	1	27 June 2017
Summary CV for Chief Investigator (CI) [Chief Investigator CV]	1	21 June 2017
Summary CV for supervisor (student research) [Jenny Limond CV]	1	23 June 2017
Validated questionnaire [HADS questionnaire]	1	04 April 2017

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document “After ethical review – guidance for researchers” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance>

We are pleased to welcome researchers and R & D staff at our RES Committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

17/LO/1185	Please quote this number on all correspondence
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With the Committee's best wishes for the success of this project.

Yours sincerely



PP Dr Anand Patel
Chair

Email: nrescommittee.london-surreyborders@nhs.net

Enclosures: *"After ethical review – guidance for researchers"*

Copy to: *Ms Gail Seymour*

*Dr Thomas Lewis, Great Ormond Street Hospital NHS Foundation
Trust & UCL ICH*

28.11.2017

Dr Jessica Watts
Trainee Clinical Psychologist
Somerset and Taunton NHS Foundation Trust

Dear Jessica

PI: Sarah Aylett

R&D number: 17BB31

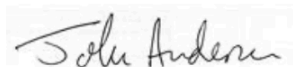
Title: Can a Short-Term Distance-Based Intervention Improve Implementation of Neuropsychological Recommendations in School-Aged Children with Sturge-Weber Syndrome?

Thank you for taking the time to speak with us today. The Committee is satisfied that any concerns have now been addressed and has no objections to the conduct of this project at

You will shortly be contacted by R&D Governance who will support you through the process of obtaining the necessary approvals before your project can begin. You must not commence your project before receiving R&D approval. Please find attached further information regarding the next stages in the research administration process.

Decision: Approval

Regards,



Professor John Anderson
Chair
Clinical Research Adoption Committee

The child first and always

Dear Jessica Watts,

Application
ID: eCLESPsy000187 v2.1

Title: An investigation into the impact of a short-term distance based intervention on the implementation of neuropsychological recommendations for families with a child who has a complex physical health condition

Your e-Ethics application has been reviewed by the CLES Psychology Ethics Committee.

The outcome of the decision is: **Favourable**

Potential Outcomes

Favourable:	The application has been granted ethical approval by the Committee. The application will be flagged as Closed in the system. To view it again, please select the tick box: View completed
Favourable, with conditions:	The application has been granted ethical approval by the Committee under the provision of certain conditions. These conditions are detailed below.
Provisional:	You have not been granted ethical approval. The application needs to be amended in light of the Committee's comments and re-submitted for Ethical review.
Unfavourable:	You have not been granted ethical approval. The application has been rejected by the Committee. The application needs to be amended in light of the Committee's comments and resubmitted / or you need to complete a new application.

Please view your application [here](#) and respond to comments as required. You can download your outcome letter by clicking on the 'PDF' button on your eEthics Dashboard.

If you have any queries please contact the CLES Psychology Ethics Chair:

Lisa Leaver L.A.Leaver@exeter.ac.uk

Kind regards,
CLES Psychology Ethics Committee

Appendix C: Information, letters and forms sent to families**Study Invitation Letter**

Date: DD/MM/YYYY

Dear (***child's name***) and parents,

We are writing to you to let you know about a study that you are eligible to participate in. Full information about the study is included with this letter, along with the consent form.

Please take time to read the information sheet and contact the chief investigator with any questions. We have included a pre-paid self-addressed envelope for you should you wish to return the consent form. If we have not received the form from you after two weeks (**insert date**) the chief investigator will be in touch to answer any questions you may have.

Thank-you for taking the time to consider the study.

Yours Sincerely,

Jessica Watts
Trainee Clinical Psychologist,
Exeter University



Participant Information Sheet (PIS)

Introduction

You are receiving this study invitation because your child (*insert child's name*) sees the Sturge-Weber and Neurocutaneous Syndrome Service (SWaNS) at XXXX.

This research is being completed to improve outcomes for families where their child has complex health needs. This research forms part of my qualification to be a clinical psychologist at University of Exeter.

What is the purpose?

This study aims to support families implement behaviour and environmental recommendations within the home.

How will the study be completed?

The study will be completed using technology, which means you do not need to travel. All aspects have been designed to be quick and easy to complete and therefore will not take a significant period of time.

There are four main phases to the study:

1. Telephone conversations to complete questionnaires and discuss your most recent clinical report with the chief investigator.
2. Regular prompts (either by text or email) to record what recommendations within the home you are been able to implement and to what extent.
3. A second telephone conversation to complete some questionnaires (there are less at this stage compared to stage 1) and talk more about the recommendations. At this stage you will be sent a visual aid to support implementing the recommendations. You will then receive the same prompts as in stage 2 to record which recommendations you are implementing.
4. Final telephone conversation to complete the final questionnaires and answer any final questions about the study.

What are the possible disadvantages?

There are no known risks to participating in this study. However, it may be that through attempting to change behaviour patterns in the home that you or your child

will experience some distress. In the unlikely event this happens, it is not anticipated to continue after the study. Should this occur or any of your circumstances change, then you are reminded that you are able to withdraw from the study at any time.

What are the possible advantages?

It is possible that through participating your family will feel more able to follow up on the recommendations provided by the team at XXXX. In turn, this could lead to a change in your child's outcomes.

Additionally, it is hoped that through your participation that researchers will have a better understanding of how best to support families implement recommendations.

Do I have to take part?

Your participation is completely voluntary. This means that you do not have to take part now and also that if you change your mind partway through the study you are able to withdraw, without providing any reason. The study is also entirely separate to the service provided within the NHS and at XXXX therefore your participation will not influence or affect any of the care you receive now or in the future.

What about my confidentiality?

Your confidentiality will be respected, which means that other than the researchers, no-one will have access to your responses. All files containing any of your information (or responses) will be stored on a password-protected file on a password-protected computer, which only the researchers can access. You will be able to contact the researchers at any time during the study if you have any questions.

What happens after the research?

The researchers will share the findings with the clinical team and aim to publish the results. These results will be anonymised, and your information will not be identifiable.

Other information

The study has been independently reviewed and given a favourable opinion by the XXXX NHS Research Ethics Committee.

Should you wish to complain about any aspect of the study please first contact the chief investigator or research supervisor using the details below. However, if you would like to take any concerns further, please contact the research sponsor Gail Seymour, by email g.m.seymour@exeter.ac.uk or phone, 01392 726 621.

Contact details

Chief Investigator:
Jessica Watts,
Trainee Clinical Psychologist,
Exeter University
Jw735@exeter.ac.uk



Clinical lead:

Research Supervisor:
Dr Jenny Limond
Consultant Paediatric Neuropsychologist and Senior Lecturer at Exeter University
J.Limond@exeter.ac.uk

**Child Participant Information Sheet (PIS)****Introduction**

You are being invited to take part in a study as you see some doctors at XXXX.

Why?

The study wants to help you and your family with some of doctor's suggestions. This might help you with some of the things that you want to do – which might be to be more independent or 'grown up' or maybe to not get cross or upset with your family.

How?

You won't need to visit the hospital or do any 'tests'. We can talk on the phone or by video. I will also talk to your Mum or Dad on the telephone and maybe with email or text messages too. You will have three chances to speak to me on the phone if you want but you can choose if you would like to speak with me or not.

What could happen?

Taking part in the study, might help you with some of the things you find difficult. You might also find it hard to do some of the things we talk about and this might make you or your family a bit upset at times. This feeling shouldn't last long, and you can always stop the study whenever you and your parents want.

Do I have to?

No. You and your family can change your mind at any time. But it might be a good idea to talk to your parents about why you don't want to take part any more.

What happens after?

Some of the findings from this study might be useful to other doctors and we will talk about the study with them. We will keep your information private from anyone who is not involved in the study, and your name will not appear in any study results.

Contact details

Chief Investigator:
Jessica Watts,
Trainee Clinical Psychologist,
Exeter University
Jw735@exeter.ac.uk



Clinical lead:

Research Supervisor:
Dr Jenny Limond
Consultant Paediatric Neuropsychologist and Senior Lecturer at Exeter University
J.Limond@exeter.ac.uk



Informed Consent Sheet

I, the undersigned, confirm that (please tick box as appropriate):

1.	I have read and understood the information about the project, as provided in the Participant Information Sheet (version 1)	<input type="checkbox"/>
2.	I have been given the opportunity to ask questions about the project and my participation.	<input type="checkbox"/>
3.	I voluntarily agree to participate in the project.	<input type="checkbox"/>
4.	The study has been explained to my child by myself (the parent) and / or the researcher. My child has been able to ask questions and they are happy to participate.	<input type="checkbox"/>
5.	I understand I can withdraw myself and my child at any time without giving reasons. I understand that I will not be penalised for withdrawing nor will I be questioned on why I have withdrawn.	<input type="checkbox"/>
6.	I understand that my decision to withdraw will not affect the care of my child at XXXX now or in the future.	<input type="checkbox"/>
7.	The procedures regarding confidentiality have been clearly explained (e.g. anonymisation of data, etc.) to me.	<input type="checkbox"/>
8.	I consent for the researcher to access my child's medical record in relation to this study.	<input type="checkbox"/>
9.	I, along with the chief investigator, agree to sign and date this informed consent form.	<input type="checkbox"/>

Parent:

Name of Parent

Signature

Date

Name of Child

Child's signature

Date

Researcher:

Jessica Watts

Name of Researcher

Signature

Date

**Child assent form**

(to be completed by child and parent / guardian)

Study to improve implementation of paediatric recommendations**Child** (or parent on their behalf) circle yes or no for following statements:

- | | |
|---|----------|
| 1. Do you understand what the project is about? | Yes / No |
| 2. Have you asked all the questions you want? | Yes / No |
| 3. Have your questions been answered in a way you understand? | Yes / No |
| 4. Are you happy to take part? | Yes / No |

If any answers are 'no' or you are not sure, then don't sign your name

If you **do** want to take part, write your name below

Name:

Date:

The researcher who explained the research has signed this form too:

Name:

Date:

Signature

Thank you for your help



Debrief Sheet

Dear (child and parent's name),

Thank-you for your time and participation in this study. As you know the study aimed to develop a simple method to help clinicians support families implement the recommendations provided in your report.

Your time and responses have been invaluable. Now the study is complete the researcher will continue to analyse the findings and ideally these will be published to the wider scientific community. The findings will also be shared with the clinical team at XXXX. As discussed before all analyses and publications will be anonymous and therefore all of your identifiable information will have been removed.

If you have any questions please do not hesitate to get in touch with the Chief Investigator. Thank-you again for your time and commitment, we hope you found it meaningful and useful.

Chief Investigator:
Jessica Watts,
Trainee Clinical Psychologist,
Exeter University
Jw735@exeter.ac.uk

Structure for initial conversation with families

General structure of initial conversations with families

N.B. Record dates and main points covered in all conversations.

Initial conversation:

1. Identify self to families and explain why families have been contacted.
2. Explain study briefly

3. Ask if families are interested in taking part (this is non-committal, but will prevent wasting the family's or researcher's time)
4. If family is happy, agree to post information and consent sheet to the family and arrange a call in two weeks time to review and answer questions.

However, if family has time - proceed to explain study in more detail. Otherwise

complete in **Second telephone conversation:**

1. Separation of research study from XXXX
 - a. Researcher is not employed by XXXX
 - b. Study participation will not affect care now or in future,
 - c. Consent can be withdrawn at any time,
 - d. Researcher cannot address questions about medical care or anything other than the neuropsychological recommendations
 - i. Researcher will always signposted back to XXXX or G.P.
2. Data collected will be stored in accordance with Data Protection Act (1998) and will be confidential. Only the researcher will have access to the one file (password protected), which links the participant number with individual.
 - a. Families can withdraw at any time and request for their data to be deleted.
3. Study related to neuropsychological recommendations (defined as those which relate to their child's behaviour or environmental needs within the home). The study is interested in whether a distance-based intervention can support families implement some of these recommendations.
 - a. Outline what participation will mean in terms of time:
 - i. Telephone or video link conversations (depending on whether family wants more time to think about study) to explain the study, screen the family, obtain consent and discuss the most recent report and family's understanding of it and the recommendations.
 - ii. Using technology (e.g. text messages or online survey) to respond to regular prompts about recommendation implementation texts for maximum of 31 days
 - iii. Final telephone or video link conversation
 - b. Family will not have to travel anywhere
 - c. Family must agree to text messages
4. Answer any questions.
5. Screen family and obtain consent (post consent sheet too, asking for it to be returned).
6. Complete data collection for demographics and experience of recent clinic visit (see below)
7. Complete RCADS, HADS, PSS, BRIEF, Conners-3 short form, PedsQL and SDQ
8. Discuss family's most recent report and collect related data see below

9. If possible speak to the child
 - a. Very simple explanation about study
 - b. Ask about their interests (favourite game, colour, best friend etc.)
 - c. If possible ask what they find hard and what they might like to get better at
 - d. Ask if can work with them and their family to work on those things
 - i. This information will be used to create the intervention tool for the family

Data collection:

Information from families:

Parents name:	
Child's name:	
Child's age (years / months):	
Child's gender:	
Child's age diagnosis:	
Frequency of appointments with health professionals about child:	
Number of other children:	
Marital status:	
Highest parental educational level:	GCSE A-Level College qualification Undergraduate degree Post-graduate degree
Employment status	Full / part time employed Self-employed

	Unemployed
	Home-maker

Possible questions or topics to guide semi-structured interview with parents:

- 1) To gauge understanding of the report and engagement with the recommendations
 - Has the family read the report...did it make sense
 - Did the family ask the clinical team any questions about once they received it
 - Did they have to wait a long time for it
 - Did the recommendations make sense and where the applicable to the family (practical?)
 - What were the recommendations about

- 2) **Pre-intervention (post-baseline)** need to identify three focused

recommendations for remainder of study

- What would you like to be easier / want support with
- Specifically ask if want support with XX recommendation
- Name the three 'targeted' recommendations

Appendix D: Questionnaires

Strengths and Difficulties Questionnaire

Strengths and Difficulties Questionnaire

P 4-17

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

Child's Name

Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, cannot stay still for long	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often complains of headaches, stomach-aches or sickness	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often has temper tantrums or hot tempers	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, tends to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally obedient, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries, often seems worried	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Helpful if someone is hurt, upset or feeling ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Constantly fidgeting or squirming	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Has at least one good friend	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often fights with other children or bullies them	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often unhappy, down-hearted or tearful	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Easily distracted, concentration wanders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Nervous or clingy in new situations, easily loses confidence	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Kind to younger children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often lies or cheats	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Picked on or bullied by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often volunteers to help others (parents, teachers, other children)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Thinks things out before acting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Steals from home, school or elsewhere	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Gets on better with adults than with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many fears, easily scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Sees tasks through to the end, good attention span	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Do you have any other comments or concerns?

Please turn over - there are a few more questions on the other side

Overall, do you think that your child has difficulties in one or more of the following areas:
emotions, concentration, behaviour or being able to get on with other people?

No	Yes- minor difficulties	Yes- definite difficulties	Yes- severe difficulties
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

If you have answered "Yes", please answer the following questions about these difficulties:

• How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

• Do the difficulties upset or distress your child?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

• Do the difficulties interfere with your child's everyday life in the following areas?

	Not at all	Only a little	Quite a lot	A great deal
HOME LIFE	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
FRIENDSHIPS	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
CLASSROOM LEARNING	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
LEISURE ACTIVITIES	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

• Do the difficulties put a burden on you or the family as a whole?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Signature

Date

Mother/Father/Other (please specify:)

Thank you very much for your help

Parental Stress Scale**Parental Stress Scale**

The following statements describe feelings and perceptions about the experience of being a parent. Think of each of the items in terms of how your relationship with your child or children typically is. Please indicate the degree to which you agree or disagree with the following items by placing the appropriate number in the space provided.

1 = Strongly disagree 2 = Disagree 3 = Undecided 4 = Agree 5 = Strongly agree

1	I am happy in my role as a parent	
2	There is little or nothing I wouldn't do for my child(ren) if it was necessary.	
3	Caring for my child(ren) sometimes takes more time and energy than I have to give.	
4	I sometimes worry whether I am doing enough for my child(ren).	
5	I feel close to my child(ren).	
6	I enjoy spending time with my child(ren).	
7	My child(ren) is an important source of affection for me.	
8	. Having child(ren) gives me a more certain and optimistic view for the future.	
9	The major source of stress in my life is my child(ren).	
10	Having child(ren) leaves little time and flexibility in my life.	
11	Having child(ren) has been a financial burden.	
12	. It is difficult to balance different responsibilities because of my child(ren).	

13	The behaviour of my child(ren) is often embarrassing or stressful to me.	
14	. If I had it to do over again, I might decide not to have child(ren).	
15	I feel overwhelmed by the responsibility of being a parent.	
16	Having child(ren) has meant having too few choices and too little control over my life.	
17	I am satisfied as a parent	
18	I find my child(ren) enjoyable	

Hospital Anxiety and Depression Scale

Hospital Anxiety and Depression Scale (HADS)

Tick the box beside the reply that is closest to how you have been feeling in the past week.
Don't take too long over you replies: your immediate is best.

D	A		D	A	
		I feel tense or 'wound up':			I feel as if I am slowed down:
	3	Most of the time	3		Nearly all the time
	2	A lot of the time	2		Very often
	1	From time to time, occasionally	1		Sometimes
	0	Not at all	0		Not at all
		I still enjoy the things I used to enjoy:			I get a sort of frightened feeling like 'butterflies' in the stomach:
0		Definitely as much	0		Not at all
1		Not quite so much	1		Occasionally
2		Only a little	2		Quite Often
3		Hardly at all	3		Very Often
		I get a sort of frightened feeling as if something awful is about to happen:			I have lost interest in my appearance:
	3	Very definitely and quite badly	3		Definitely
	2	Yes, but not too badly	2		I don't take as much care as I should
	1	A little, but it doesn't worry me	1		I may not take quite as much care
	0	Not at all	0		I take just as much care as ever
		I can laugh and see the funny side of things:			I feel restless as I have to be on the move:
0		As much as I always could	3		Very much indeed
1		Not quite so much now	2		Quite a lot
2		Definitely not so much now	1		Not very much
3		Not at all	0		Not at all
		Worrying thoughts go through my mind:			I look forward with enjoyment to things:
3		A great deal of the time	0		As much as I ever did
2		A lot of the time	1		Rather less than I used to
1		From time to time, but not too often	2		Definitely less than I used to
0		Only occasionally	3		Hardly at all
		I feel cheerful:			I get sudden feelings of panic:
3		Not at all	3		Very often indeed
2		Not often	2		Quite often
1		Sometimes	1		Not very often
0		Most of the time	0		Not at all
		I can sit at ease and feel relaxed:			I can enjoy a good book or radio or TV program:
0		Definitely	0		Often
1		Usually	1		Sometimes
2		Not Often	2		Not often
3		Not at all	3		Very seldom

Please check you have answered all the questions

Scoring:

Total score: Depression (D) _____ Anxiety (A) _____

0-7 = Normal

8-10 = Borderline abnormal (borderline case)

11-21 = Abnormal (case)

Revised Children's Anxiety and Depression Scale

RCADS

NHS ID:

Child/ Young Person's NAME:

Relationship to Child/Young Person :

Date: / / 20

Time: h m

*Please put a circle around the word that shows how often each of these things happens to your child.
There are no right or wrong answers.*

1	My child worries about things	Never	Sometimes	Often	Always
2	My child feels sad or empty	Never	Sometimes	Often	Always
3	When my child has a problem, he/she gets a funny feeling in his/her stomach	Never	Sometimes	Often	Always
4	My child worries when he/she thinks he/she has done poorly at something	Never	Sometimes	Often	Always
5	My child feels afraid of being alone at home	Never	Sometimes	Often	Always
6	Nothing is much fun for my child anymore	Never	Sometimes	Often	Always
7	My child feels scared when taking a test	Never	Sometimes	Often	Always
8	My child worries when he/she thinks someone is angry with him/her	Never	Sometimes	Often	Always
9	My child worries about being away from me	Never	Sometimes	Often	Always
10	My child is bothered by bad or silly thoughts or pictures in his/her mind	Never	Sometimes	Often	Always
11	My child has trouble sleeping	Never	Sometimes	Often	Always
12	My child worries about doing badly at school work	Never	Sometimes	Often	Always
13	My child worries that something awful will happen to someone in the family	Never	Sometimes	Often	Always
14	My child suddenly feels as if he/she can't breathe when there is no reason for this	Never	Sometimes	Often	Always
15	My child has problems with his/her appetite	Never	Sometimes	Often	Always
16	My child has to keep checking that he/she has done things right (like the switch is off, or the door is locked)	Never	Sometimes	Often	Always
17	My child feels scared to sleep on his/her own	Never	Sometimes	Often	Always
18	My child has trouble going to school in the mornings because of feeling nervous or afraid	Never	Sometimes	Often	Always
19	My child has no energy for things	Never	Sometimes	Often	Always
20	My child worries about looking foolish	Never	Sometimes	Often	Always

21	My child is tired a lot	Never	Sometimes	Often	Always
22	My child worries that bad things will happen to him/her	Never	Sometimes	Often	Always
23	My child can't seem to get bad or silly thoughts out of his/her head	Never	Sometimes	Often	Always
24	When my child has a problem, his/her heart beats really fast	Never	Sometimes	Often	Always
25	My child cannot think clearly	Never	Sometimes	Often	Always
26	My child suddenly starts to tremble or shake when there is no reason for this	Never	Sometimes	Often	Always
27	My child worries that something bad will happen to him/her	Never	Sometimes	Often	Always
28	When my child has a problem, he/she feels shaky	Never	Sometimes	Often	Always
29	My child feels worthless	Never	Sometimes	Often	Always
30	My child worries about making mistakes	Never	Sometimes	Often	Always
31	My child has to think of special thoughts (like numbers or words) to stop bad things from happening	Never	Sometimes	Often	Always
32	My child worries what other people think of him/her	Never	Sometimes	Often	Always
33	My child is afraid of being in crowded places (like shopping centers, the movies, buses, busy playgrounds)	Never	Sometimes	Often	Always
34	All of a sudden my child will feel really scared for no reason at all	Never	Sometimes	Often	Always
35	My child worries about what is going to happen	Never	Sometimes	Often	Always
36	My child suddenly becomes dizzy or faint when there is no reason for this	Never	Sometimes	Often	Always
37	My child thinks about death	Never	Sometimes	Often	Always
38	My child feels afraid if he/she have to talk in front of the class	Never	Sometimes	Often	Always
39	My child's heart suddenly starts to beat too quickly for no reason	Never	Sometimes	Often	Always
40	My child feels like he/she doesn't want to move	Never	Sometimes	Often	Always
41	My child worries that he/she will suddenly get a scared feeling when there is nothing to be afraid of	Never	Sometimes	Often	Always
42	My child has to do some things over and over again (like washing hands, cleaning, or putting things in a certain order)	Never	Sometimes	Often	Always
43	My child feels afraid that he/she will make a fool of him/herself in front of people	Never	Sometimes	Often	Always
44	My child has to do some things in just the right way to stop bad things from happening	Never	Sometimes	Often	Always
45	My child worries when in bed at night	Never	Sometimes	Often	Always
46	My child would feel scared if he/she had to stay away from home overnight	Never	Sometimes	Often	Always
47	My child feels restless	Never	Sometimes	Often	Always

Appendix E: Characterisation details

Table 7

Participant characterisation information

Participant	Questionnaires				
	Raw RCADS (T-score)			HADS Score (clinical range)	
	Total	Depression	Anxiety	Depression	Anxiety
1	-	-	-	6 (normal)	10 (borderline)
2	41 (61)	8 (62)	33 (60)	2 (normal)	8 (borderline)
3	10 (40)	3 (48)	7 (39)	13 (abnormal)	7 (normal)
4	-	-	-	5 (normal)	13 (abnormal)

Key: RCADS – Revised Children’s Anxiety and Depression Scale; HADS – Hospital Anxiety and Depression Scale; FSIQ – Full Scale Intelligence Quotient; NA – Not Applicable. N.B. T-scores are calculated depending on gender and age. T-scores of 50 are considered the ‘average’ with a score of 10 points from this (above or below) being 1 standard deviation. T-scores of >65 are considered ‘normal’ for the RCADS. FSIQ scores are also calculated for age and gender and scores between 86 and 115 are considered ‘average’ and scores between 70 and 85 are considered ‘borderline’.

Abnormal or borderline scores were discussed with the parents and no further action was required.

Appendix F: Example poster

Click to add title

Any picture likes can go here..

I enjoy:

1. Kickboxing 
2. Ice-skating 
3. Playing with friends 

I would like to be:

1. More independent 
1. More confident

So I would like to:

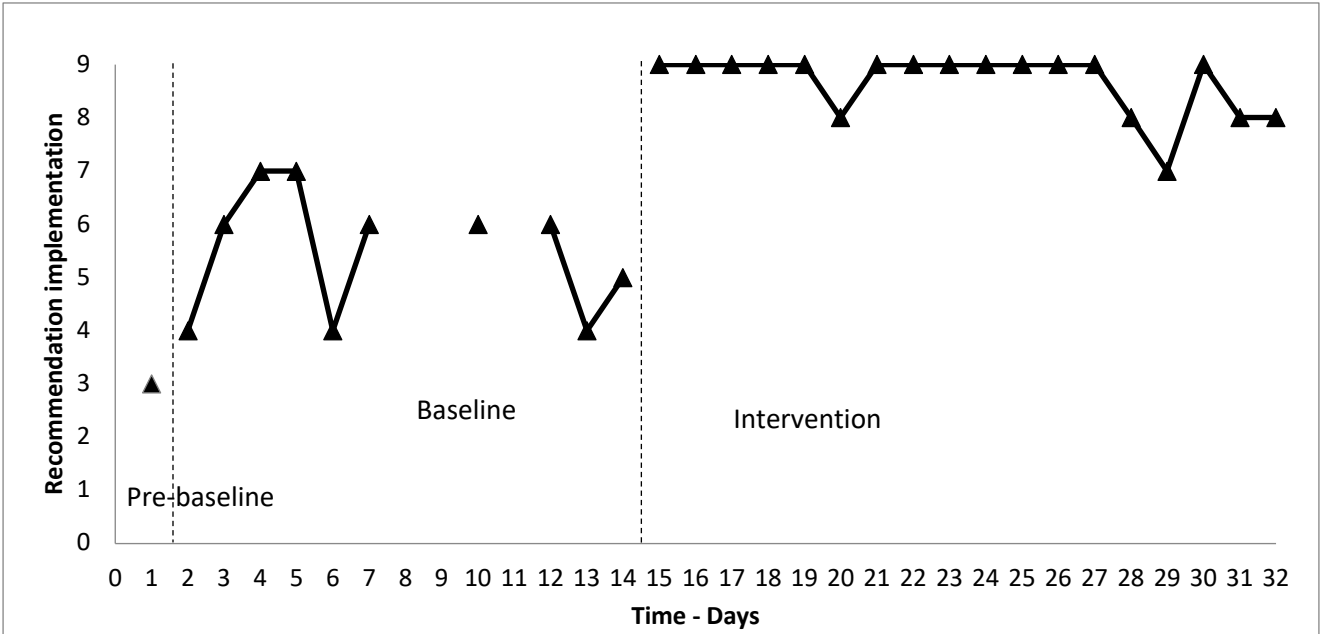
1. Read more jokes
 1. Chose a 'joke a day' from the book
 2. Put it in powerpoint
2. Walk the dog three times a week
 1. Normally after tea on Monday, Wednesday and Friday
 2. Some weeks it might be a different day, but this is OK
3. Help Mum clear the table after tea
 1. Put plates in kitchen
 2. Load the dishwasher
 3. Wipe the table too

Appendix G: Visual Analysis

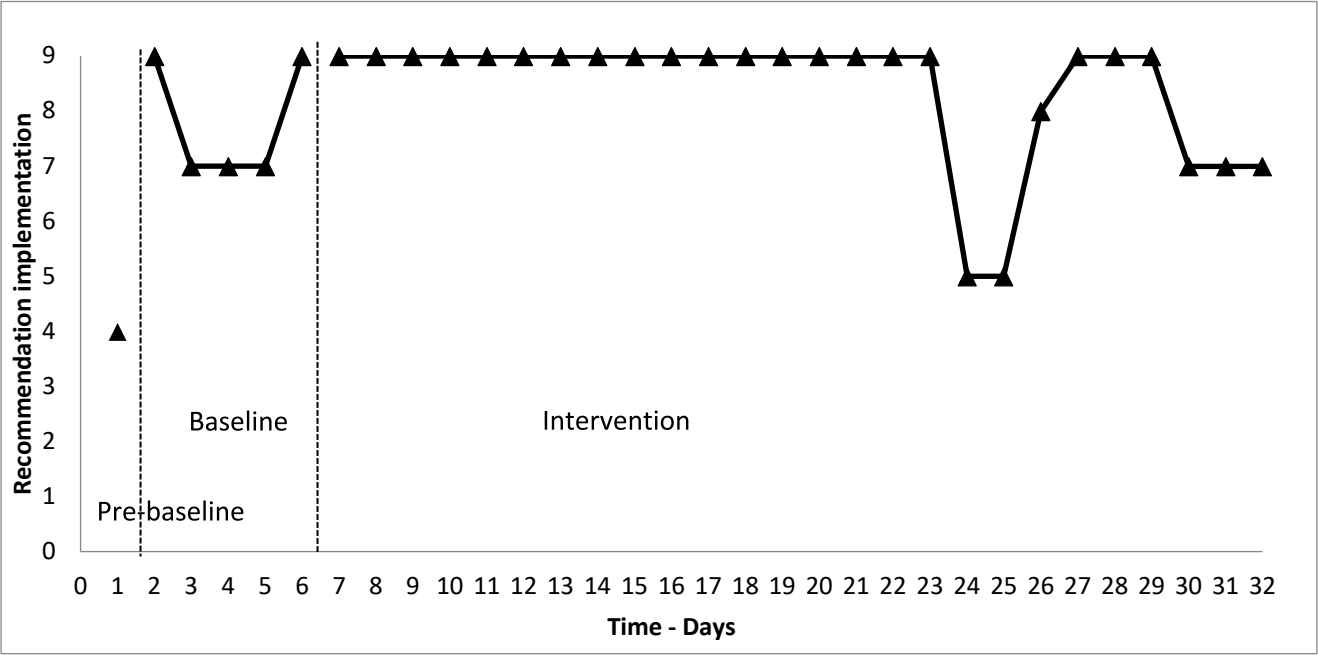
Figures for Implementation of Targeted Recommendations

Figure 4

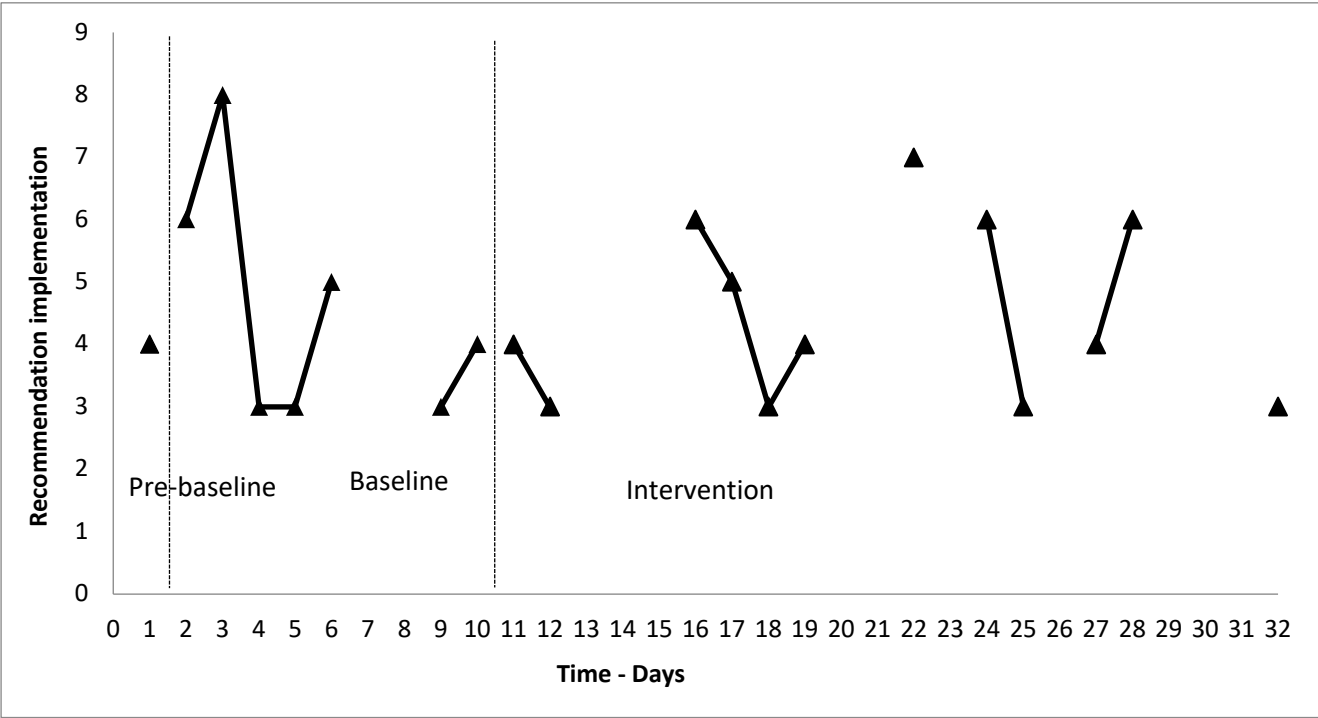
Implementation of targeted recommendations raw data (missing data visible) for all participants



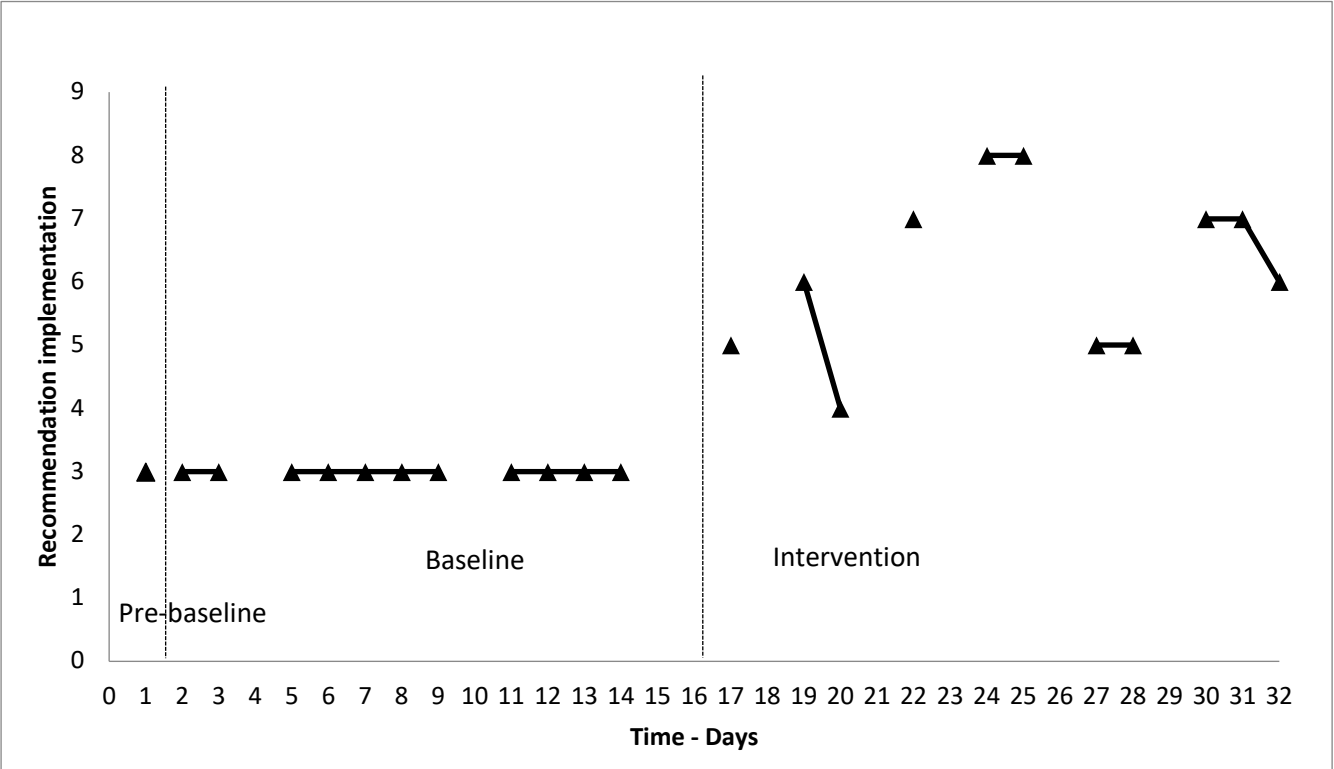
Participant 1: Implementation of targeted recommendations raw data (missing data shown as gaps)



Participant 2: Implementation of targeted recommendations raw data (no missing data)



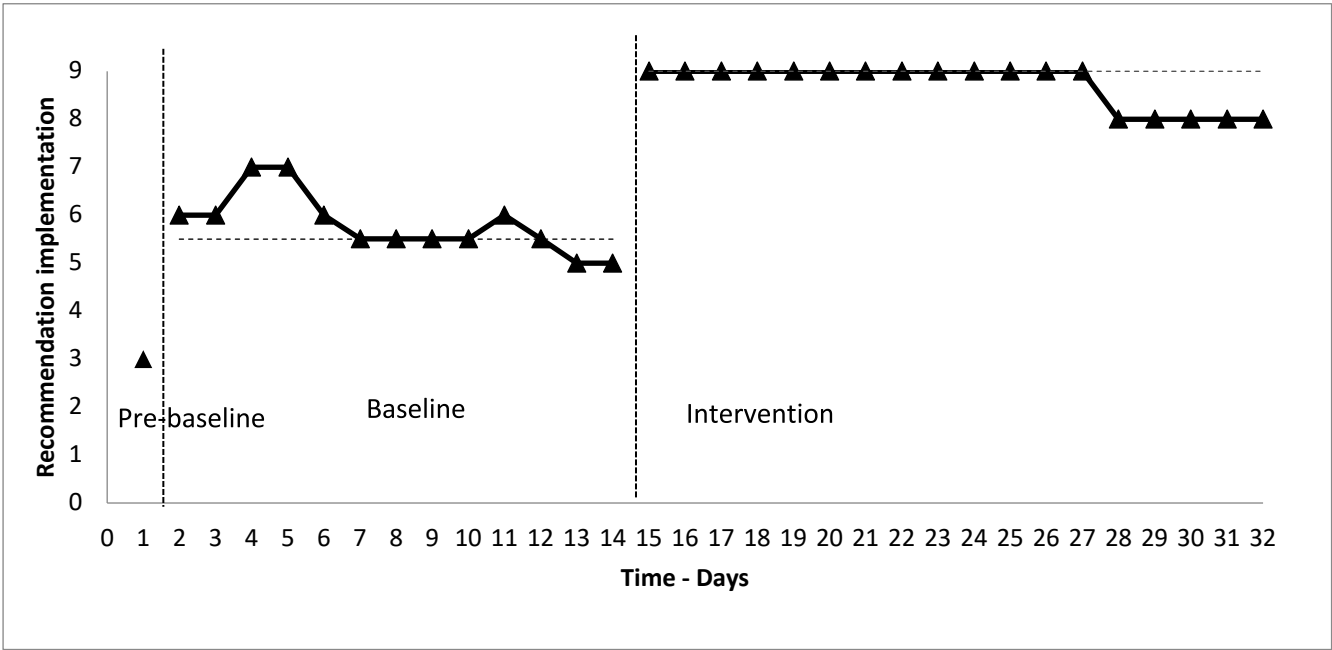
Participant 3: Implementation of targeted recommendations raw data (missing data shown as gaps)



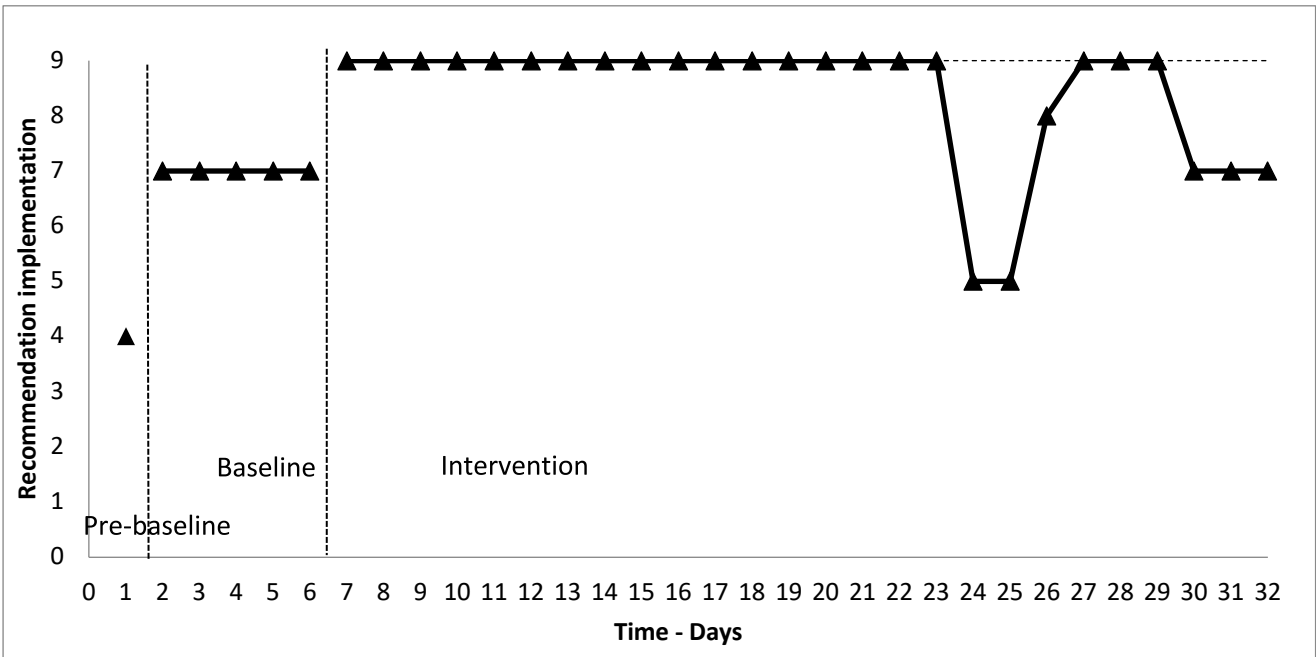
Participant 4: Implementation of targeted recommendations raw data (missing data shown as gaps)

Figure 5

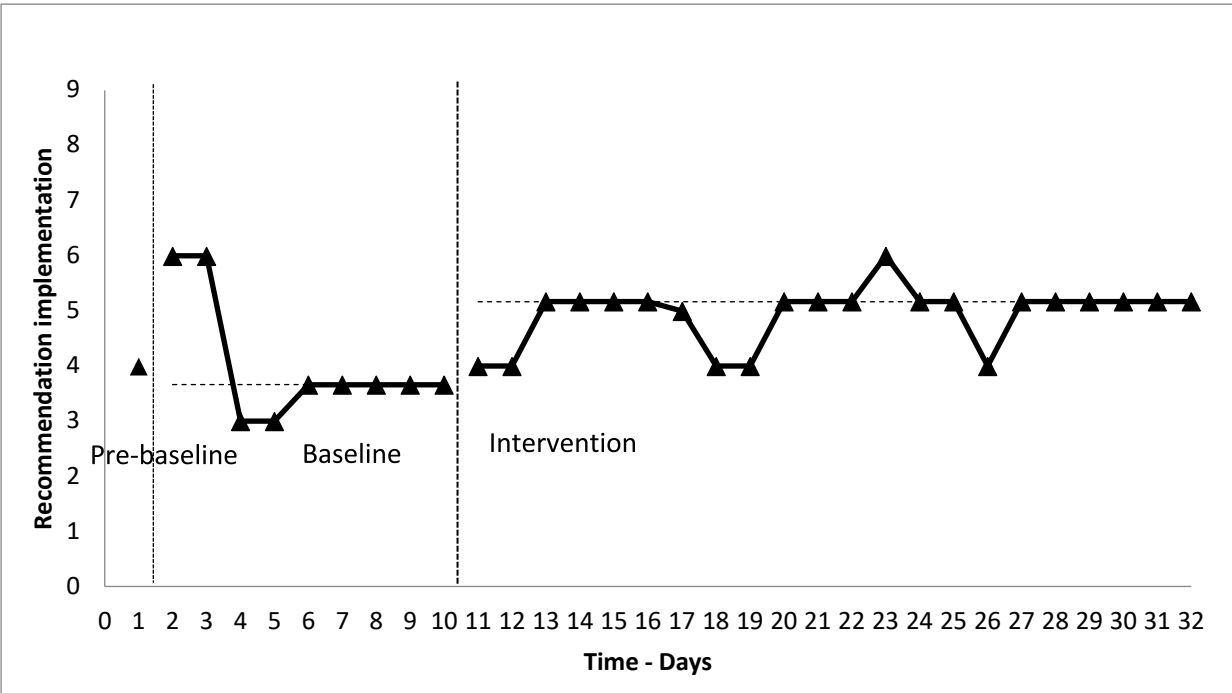
Measure of central tendency (broadened median) for all participants for targeted recommendations



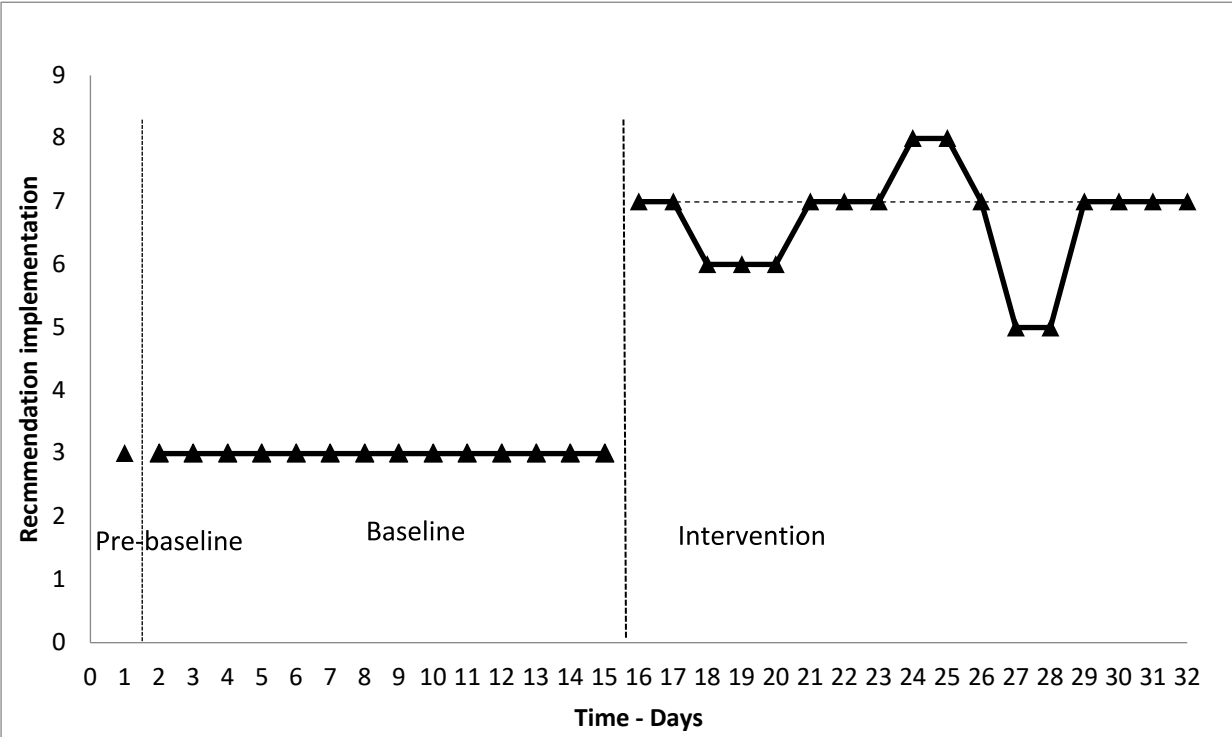
Participant 1: Median implementation for targeted recommendations



Participant 2: Median implementation for targeted recommendations



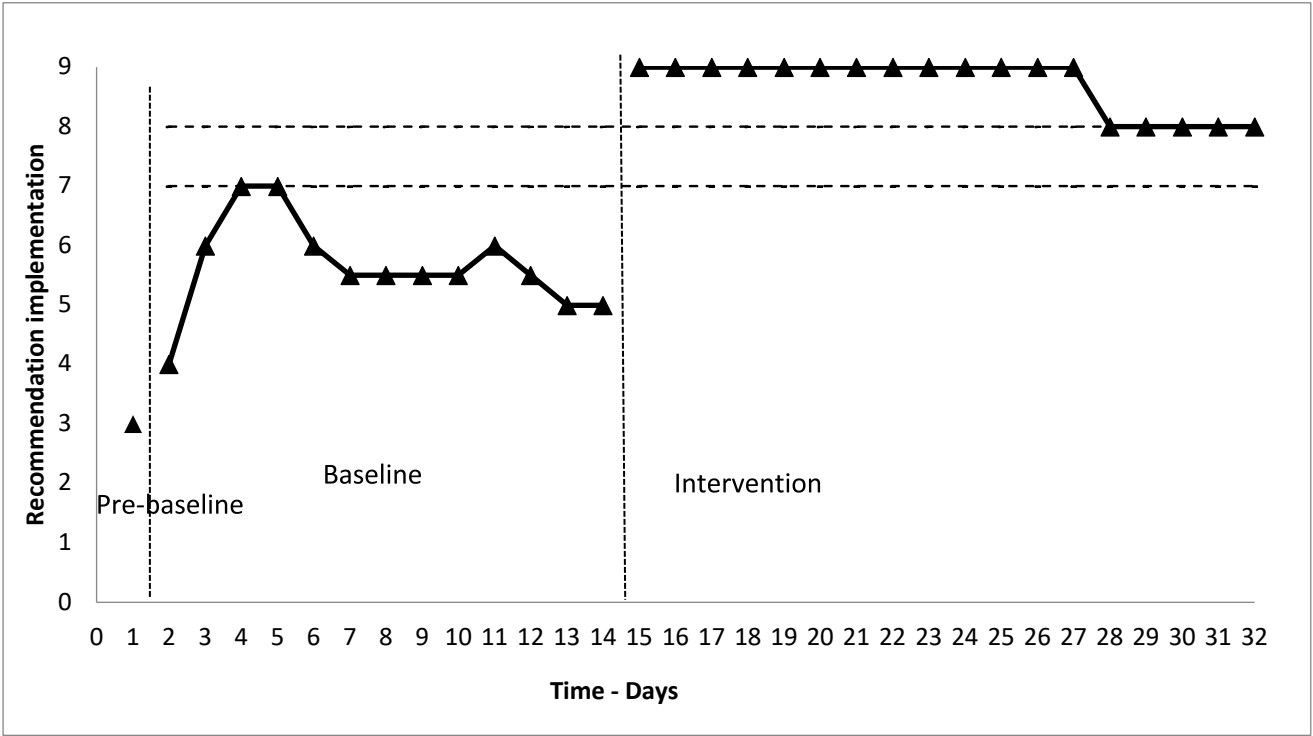
Participant 3: Median implementation for targeted recommendations



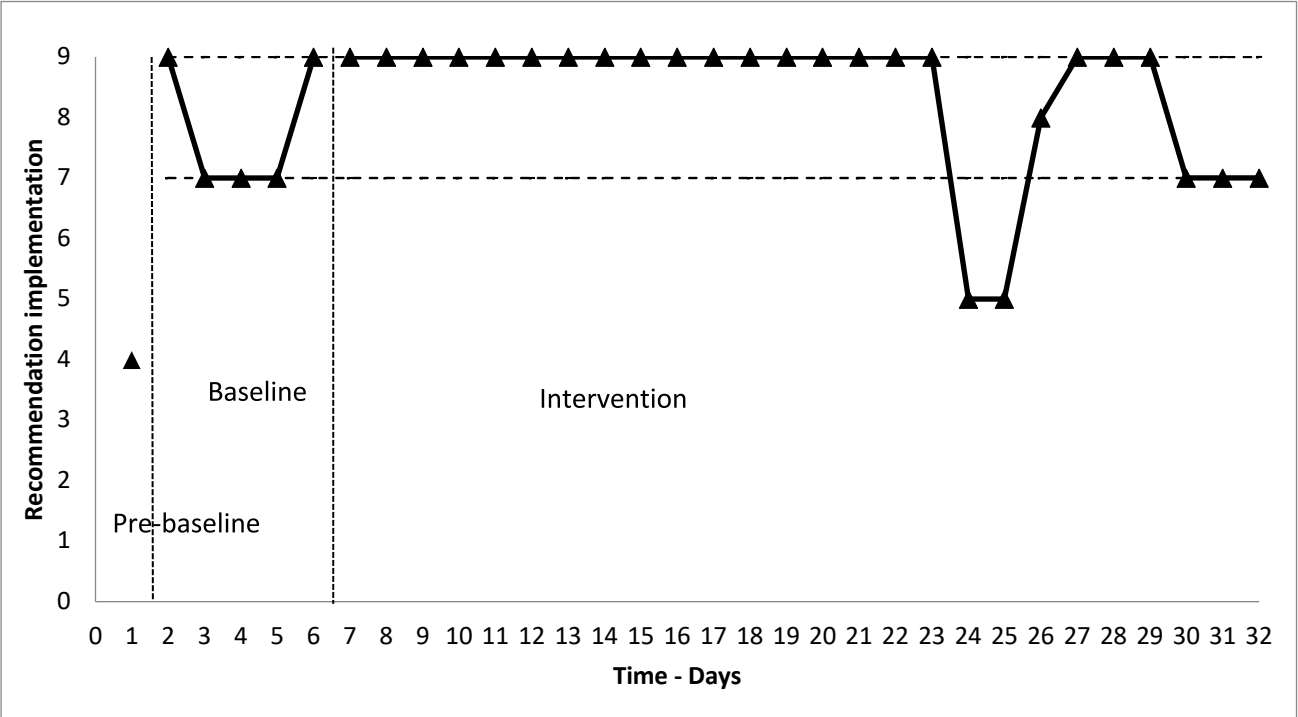
Participant 4: Median implementation for targeted recommendations

Figure 6

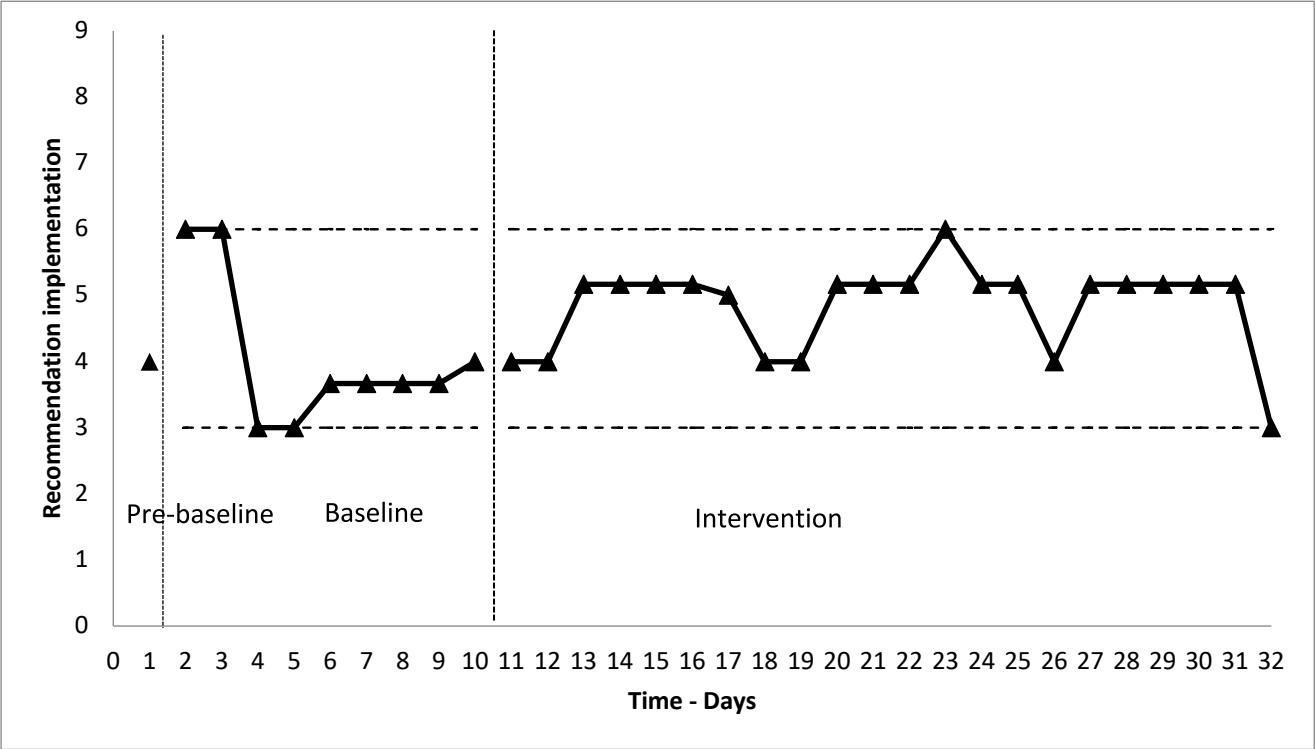
Overlap of data between phases for all participants for targeted recommendations.



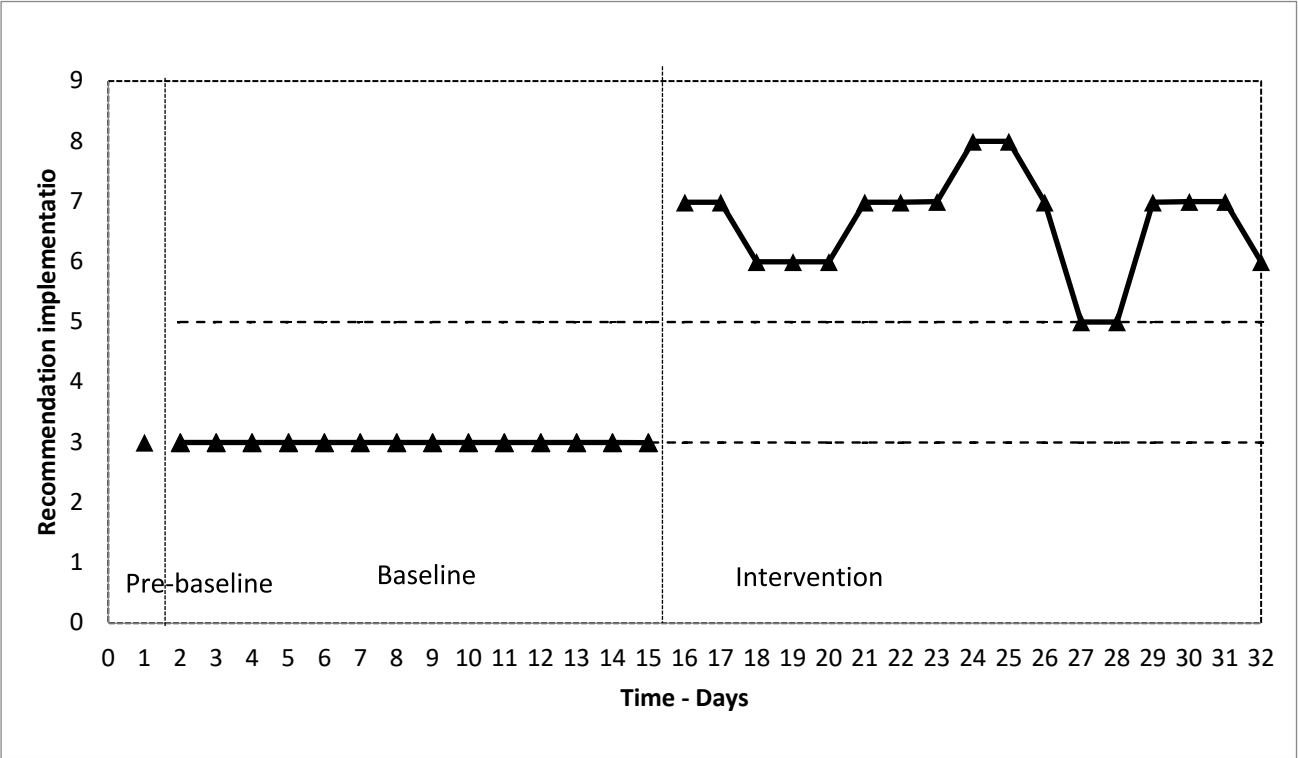
Participant 1: Overlap of data between phases for targeted recommendations



Participant 2: Overlap of data between phases for targeted recommendations



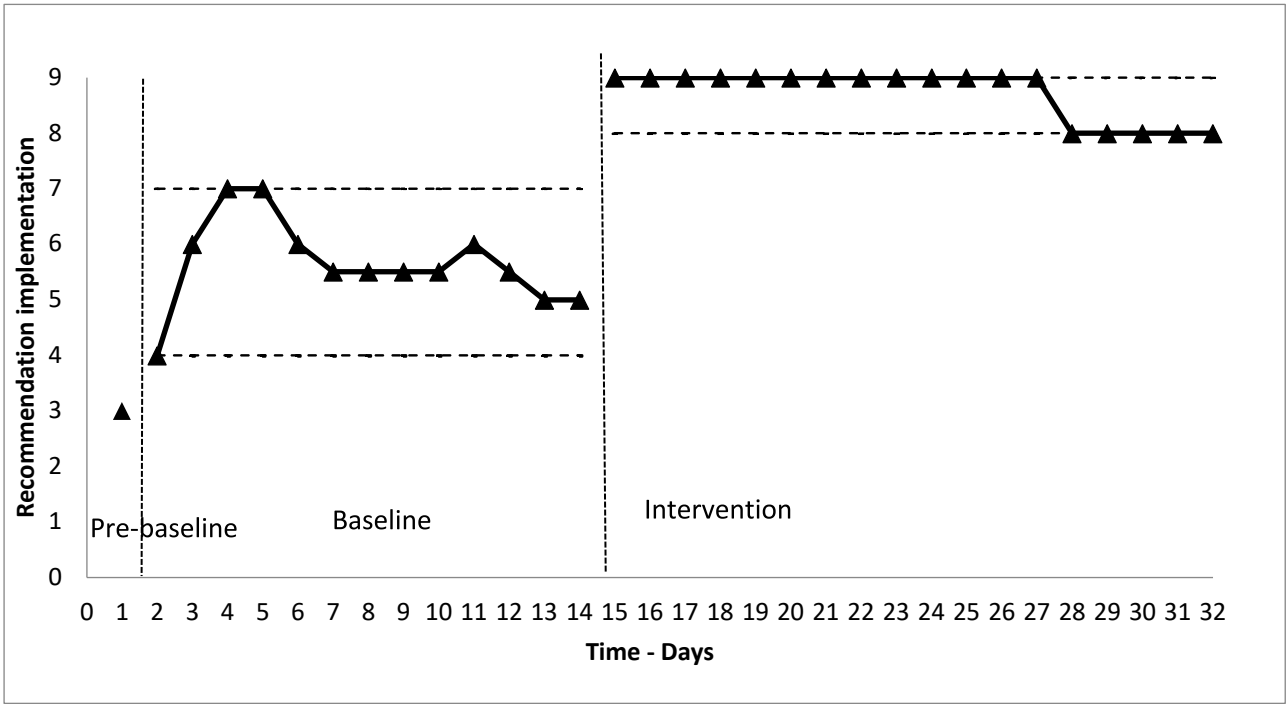
Participant 3: Overlap of data between phases for targeted recommendations



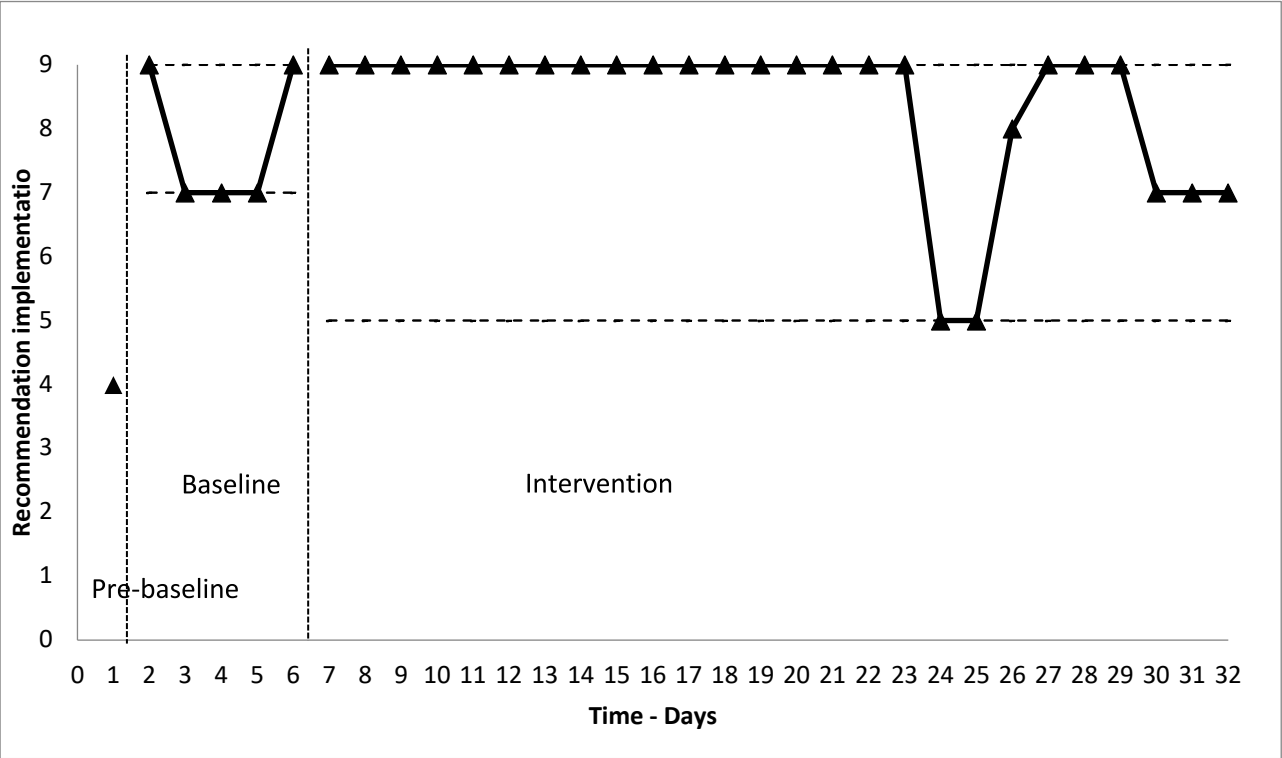
Participant 4: Overlap of data between phases for targeted recommendations

Figure 7

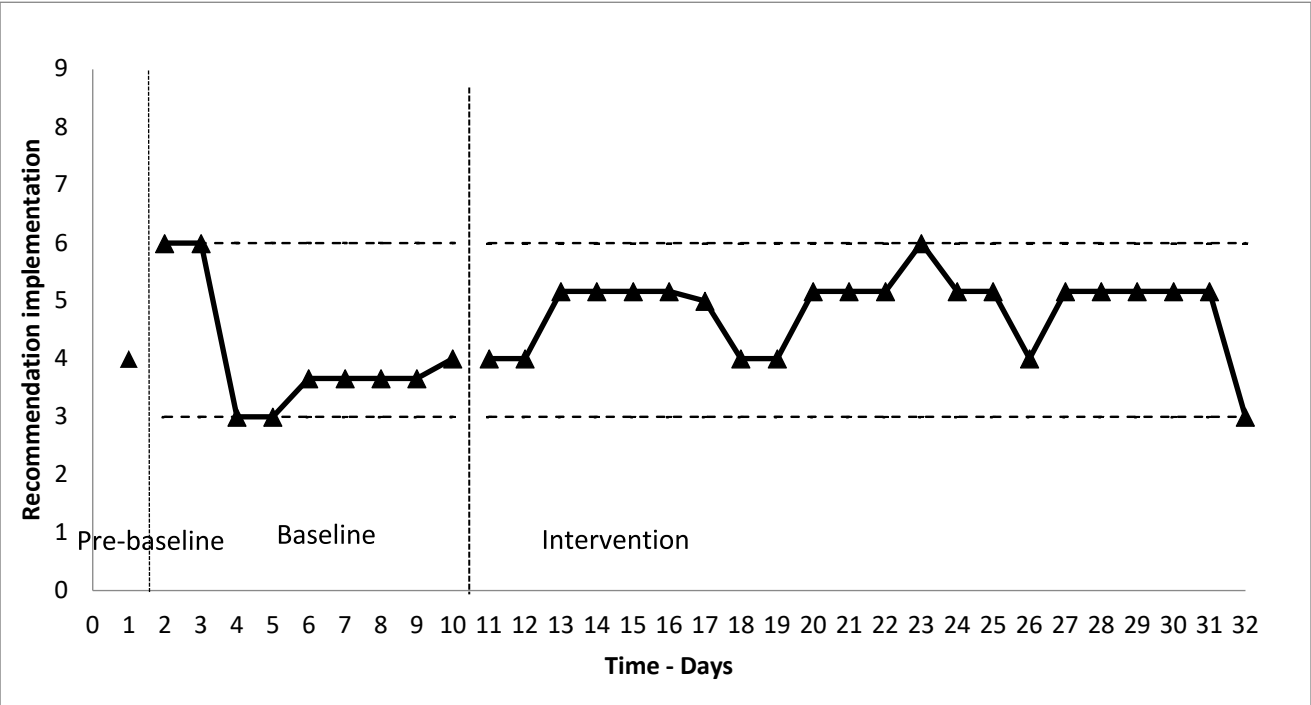
Implementation range across targeted recommendations for all participants



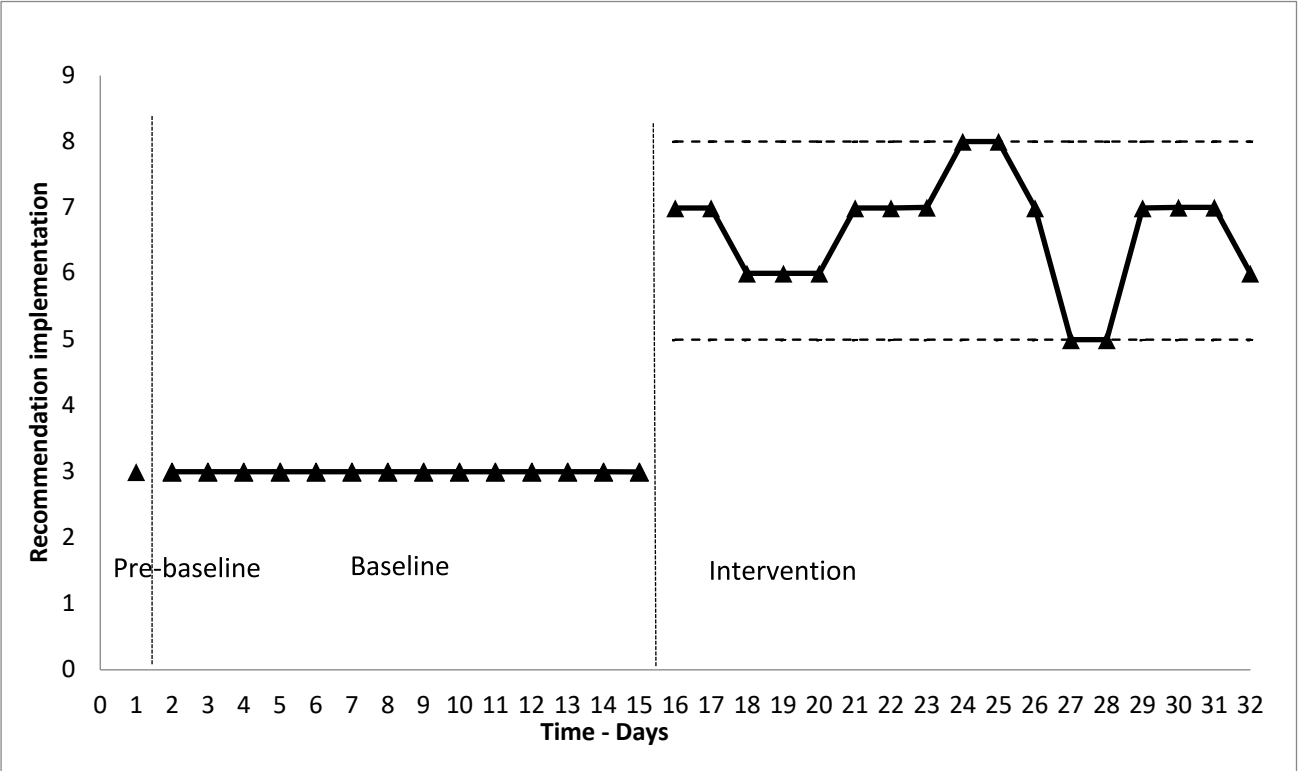
Participant 1: Implementation range across targeted recommendations



Participant 2: Implementation range across targeted recommendations



Participant 3: Implementation range across targeted recommendations

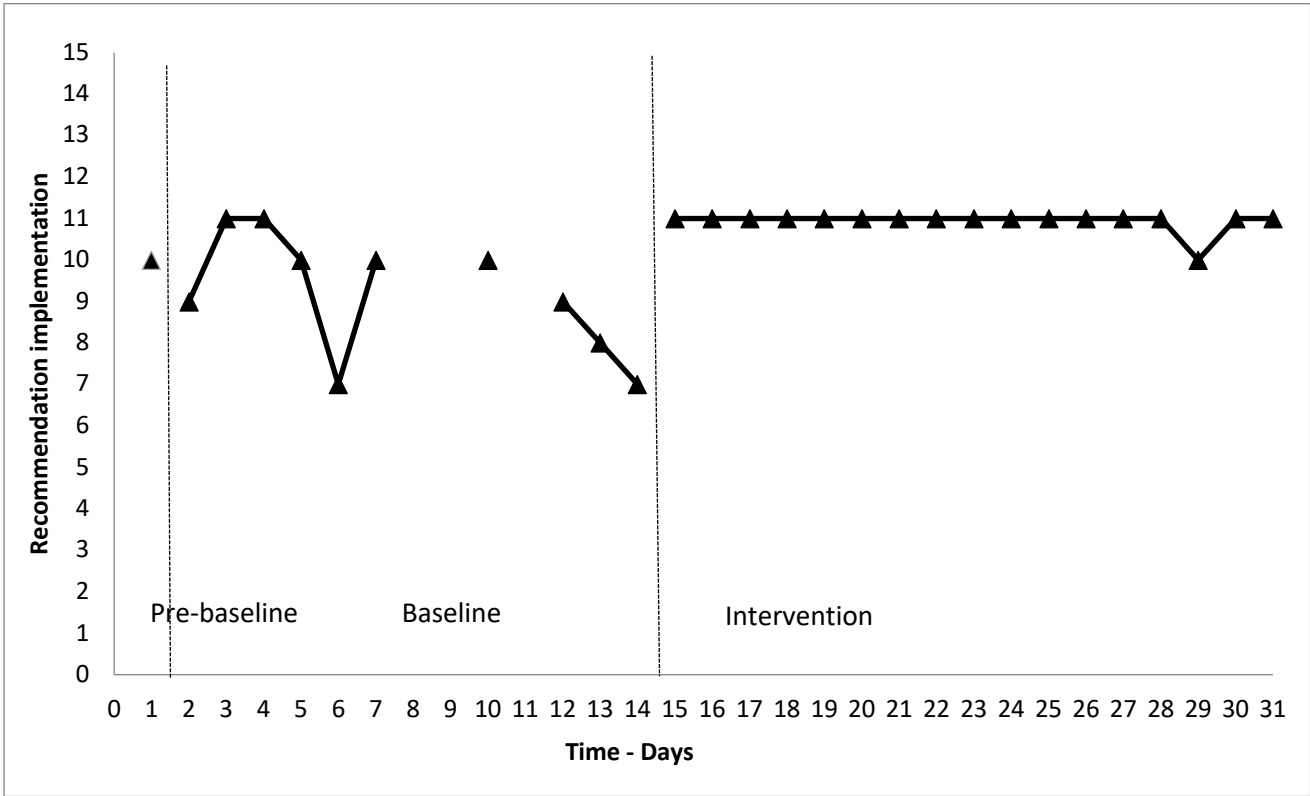


Participant 4: Implementation range across targeted recommendations

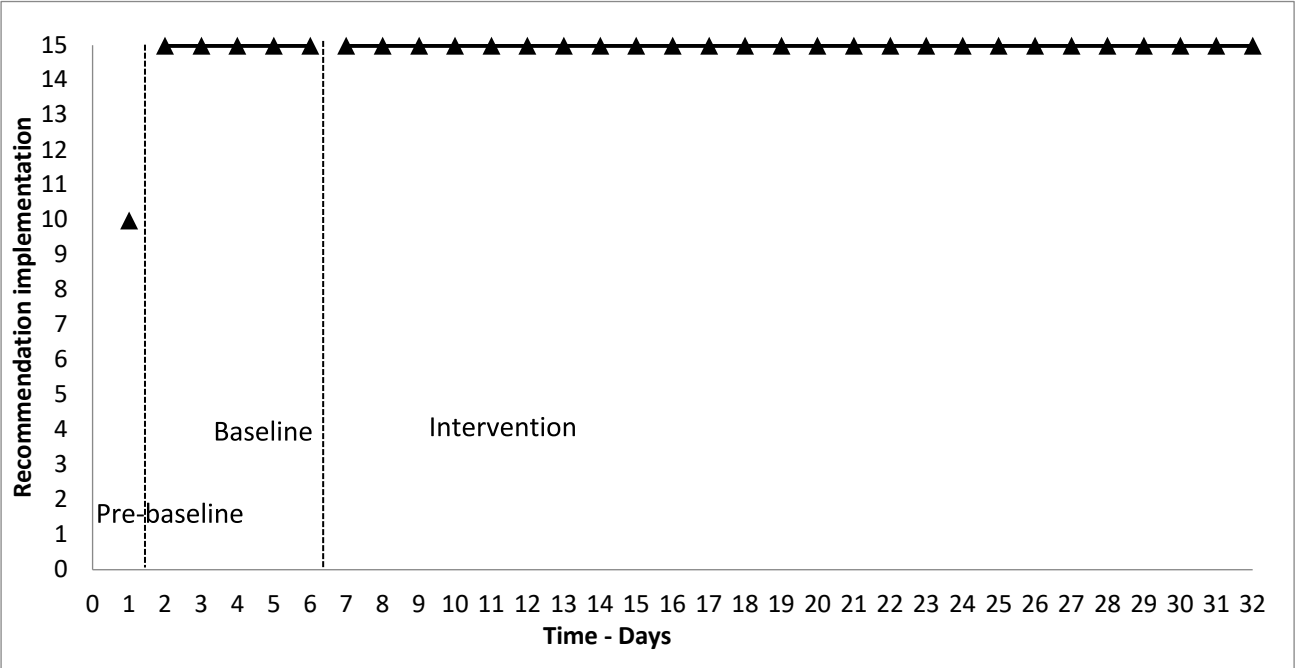
Figures for Implementation of Non-targeted Recommendations

Figure 8

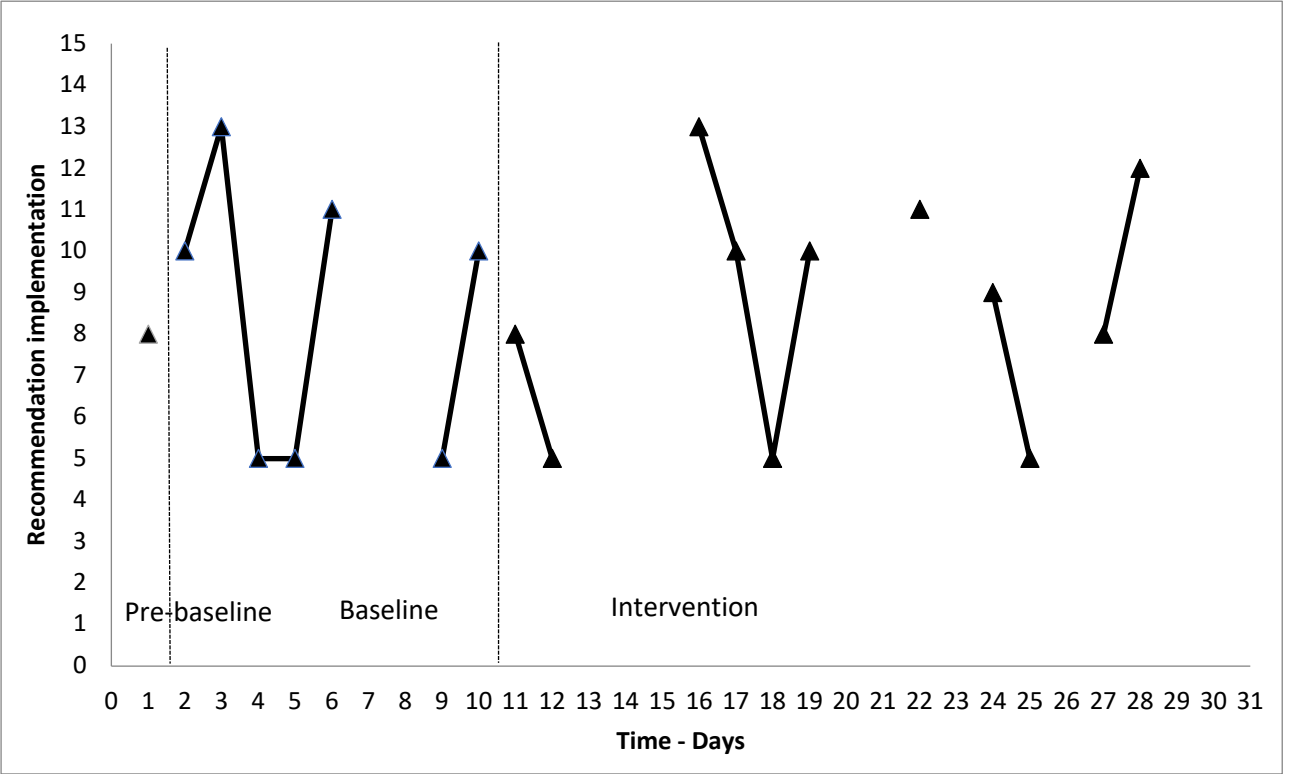
Implementation of non-targeted recommendations raw data (missing data visible) for all participants



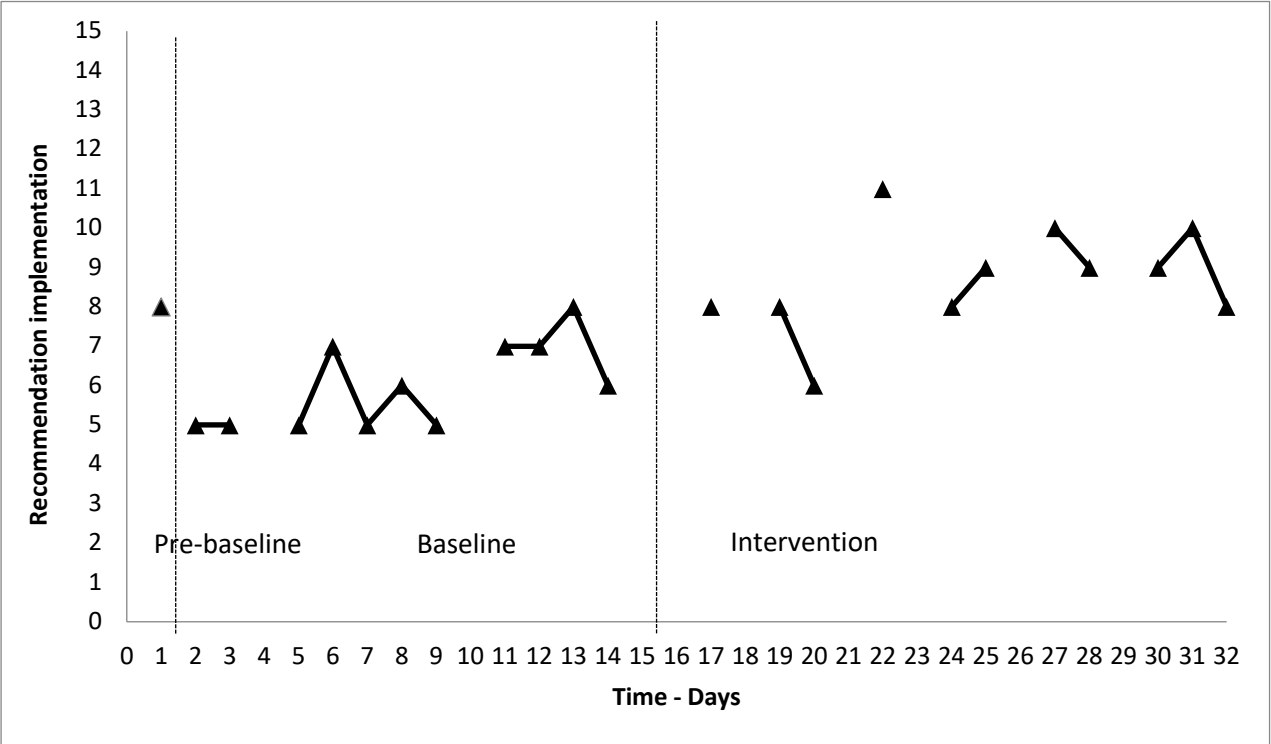
Participant 1: Implementation of non-targeted recommendations raw data (missing data shown as gaps)



Participant 2: Implementation of non-targeted recommendations raw data (no missing data)



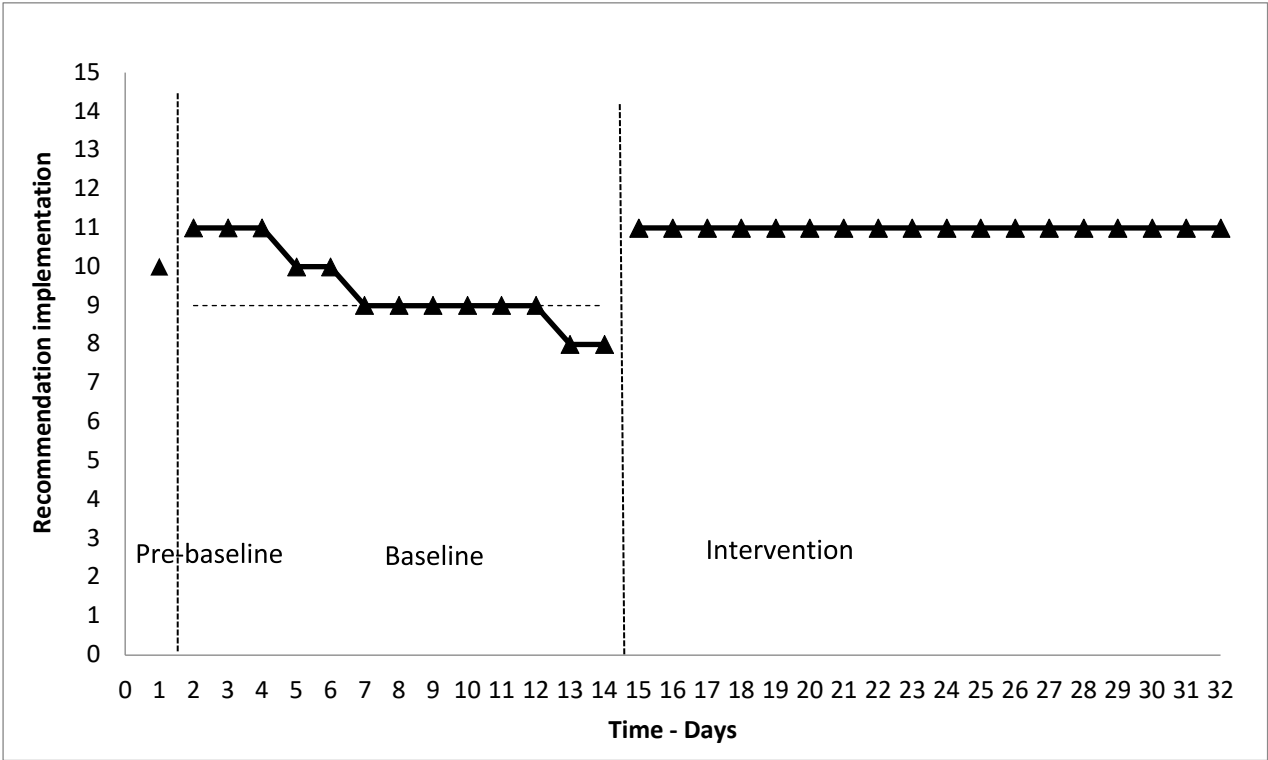
Participant 3: Implementation of non-targeted recommendations raw data (missing data shown as gaps)



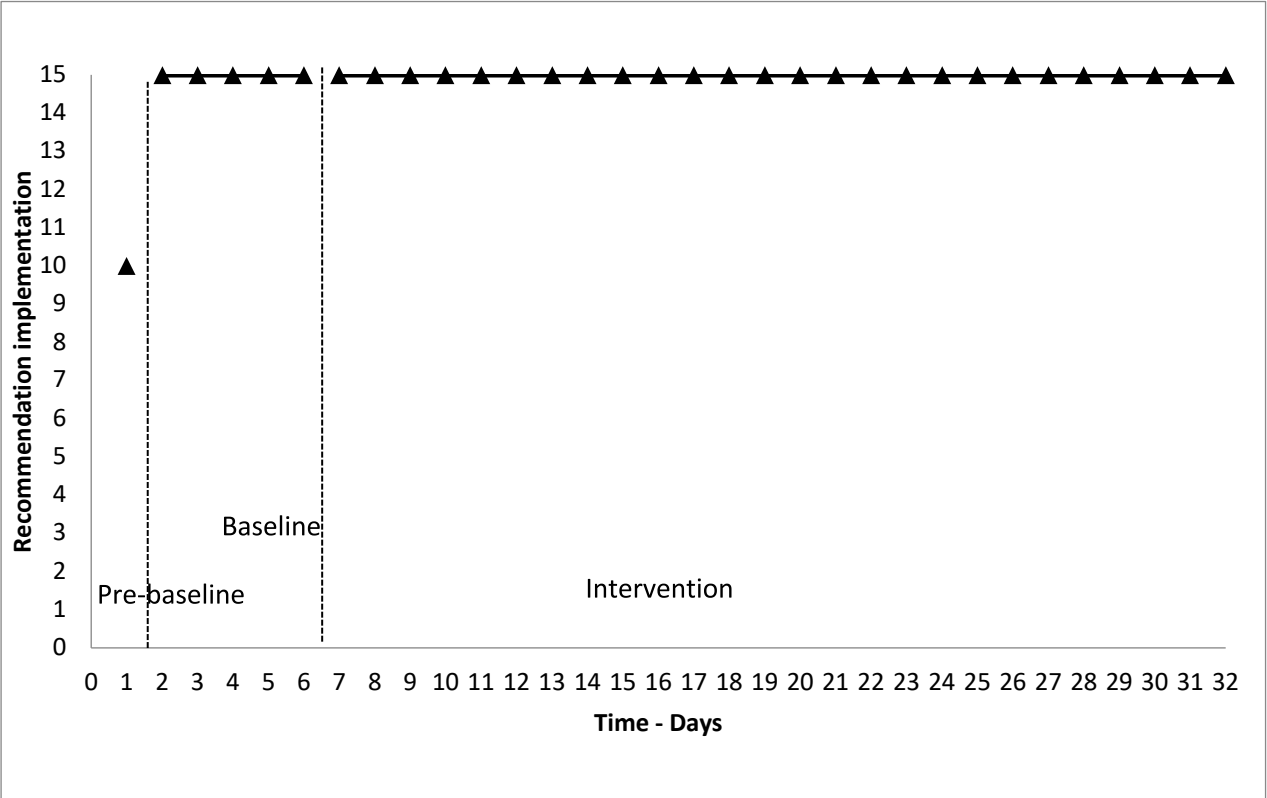
Participant 4: Implementation of non-targeted recommendations raw data (missing data shown as gaps)

Figure 9

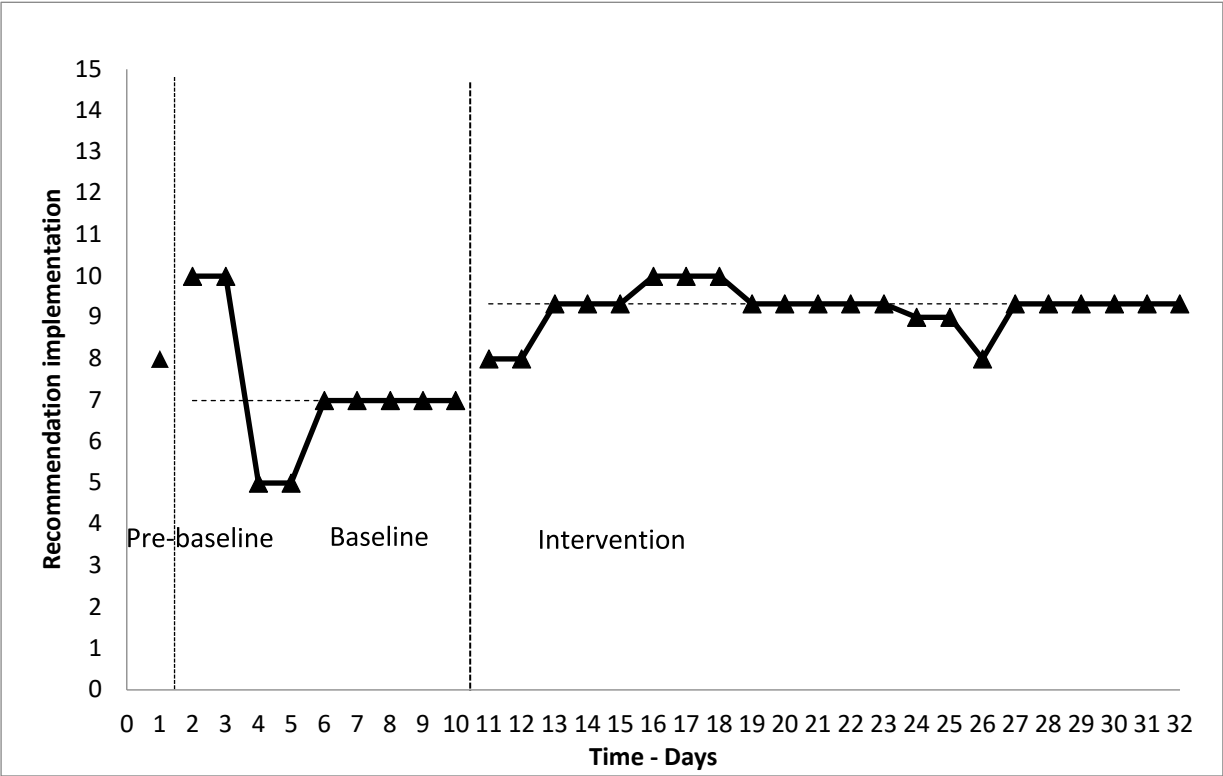
Measures of central tendency (broadened median) for all participants for non-targeted recommendations.



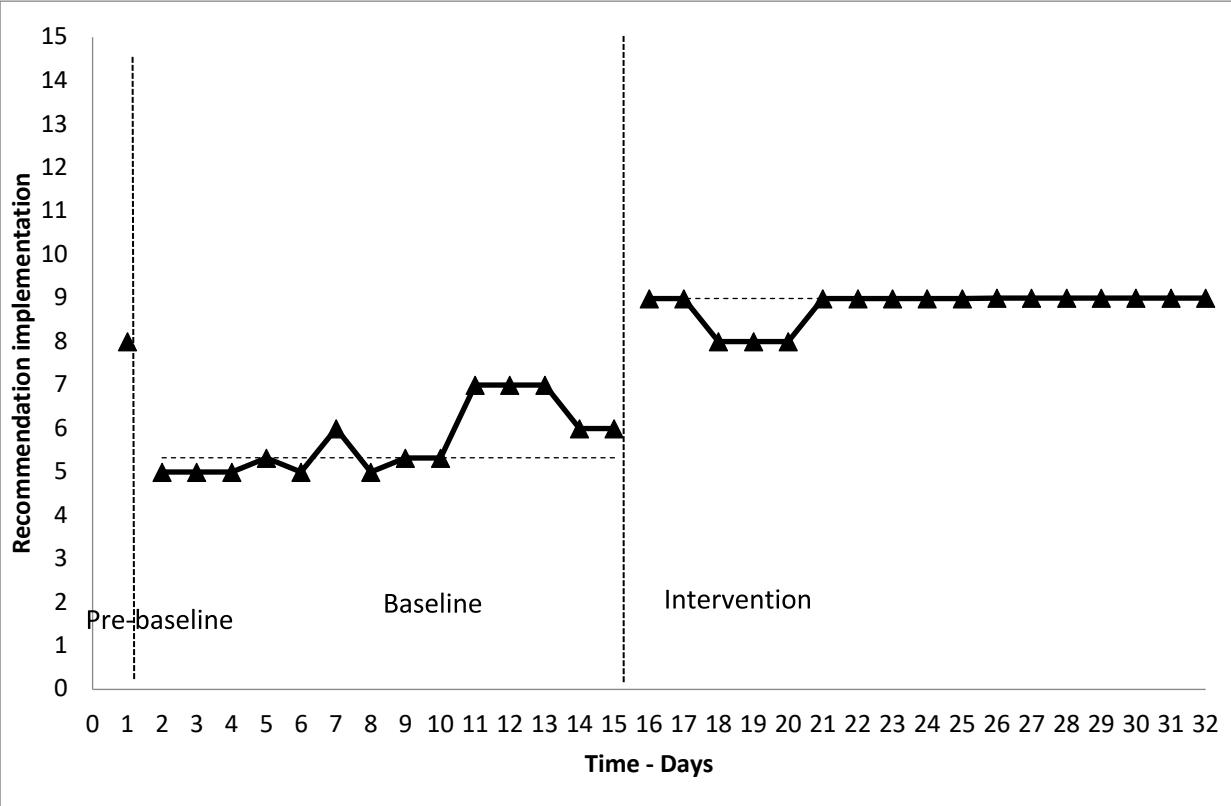
Participant 1: Median implementation for non-targeted recommendations



Participant 2: Median implementation for non-targeted recommendations



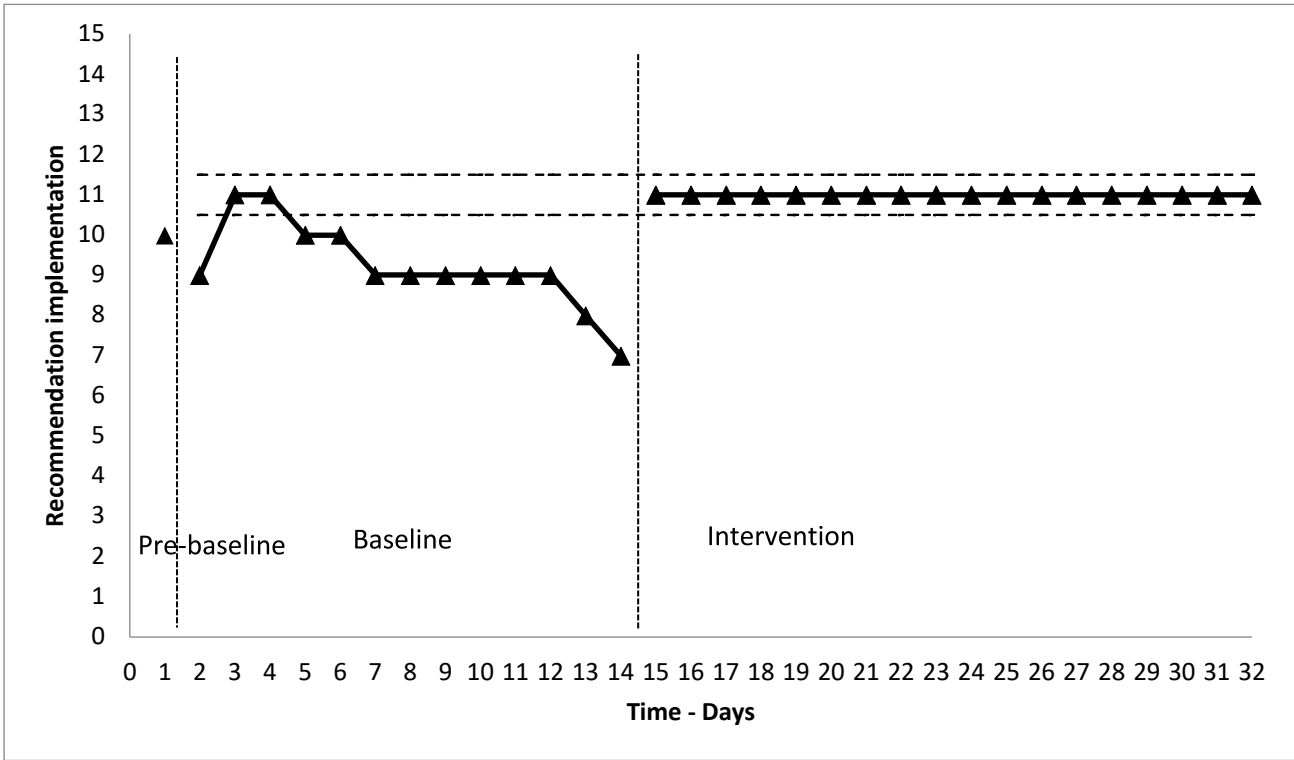
Participant 3: Median implementation for non-targeted recommendations



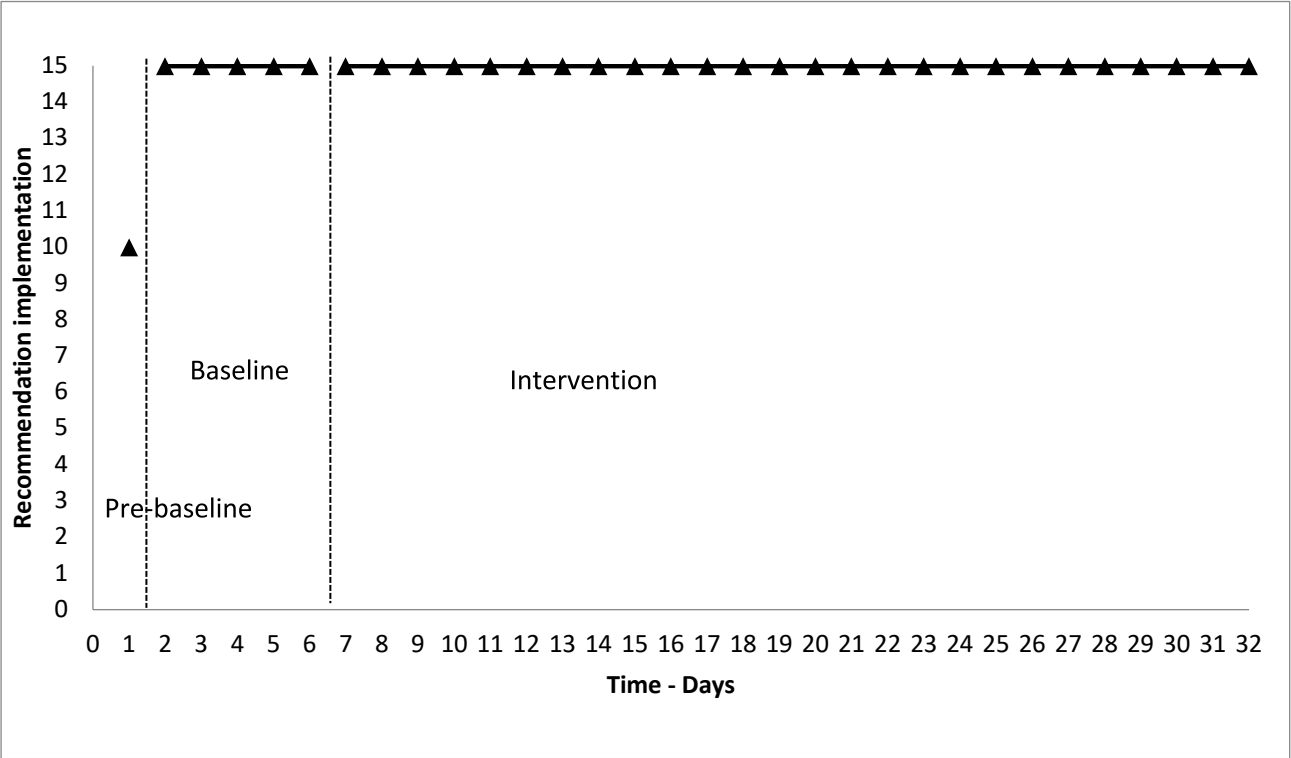
Participant 4: Median implementation for non-targeted recommendations

Figure 10

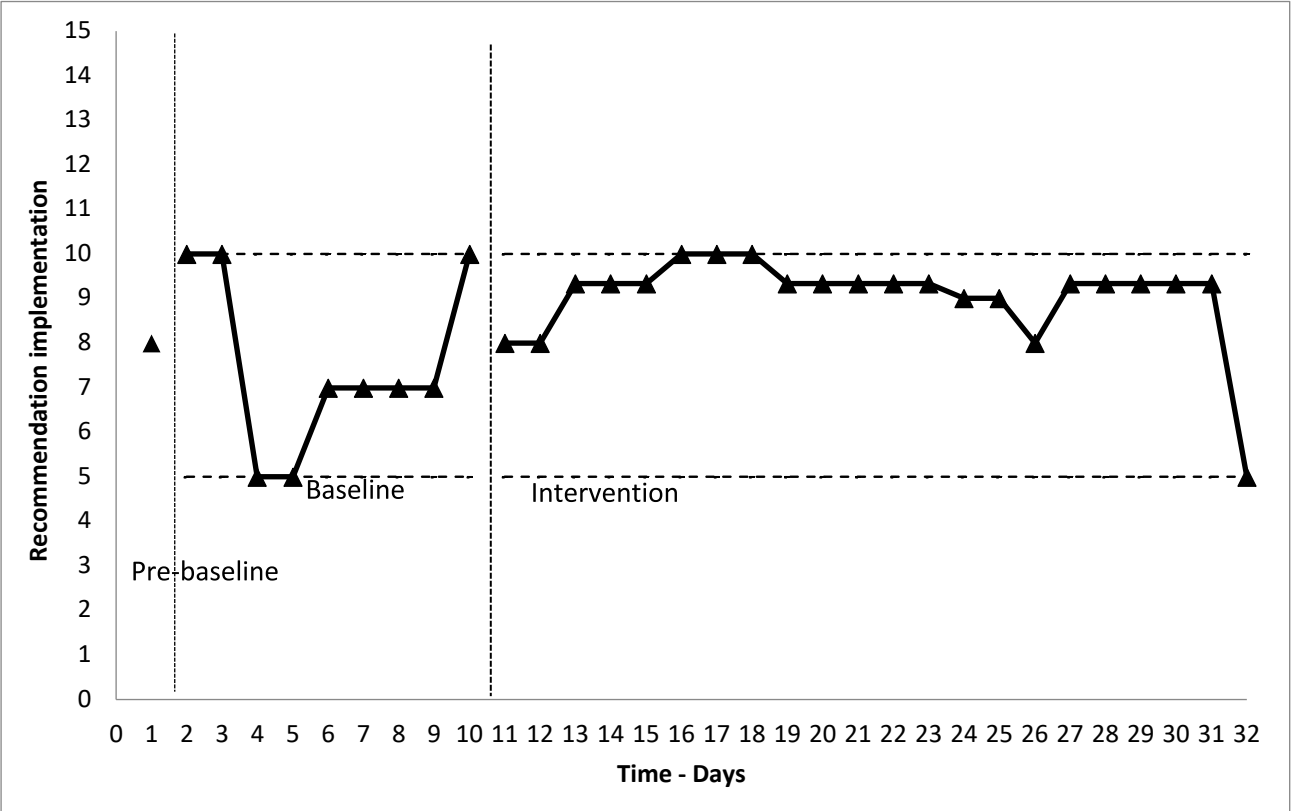
Overlap of data between phases for non-targeted recommendations for all participants



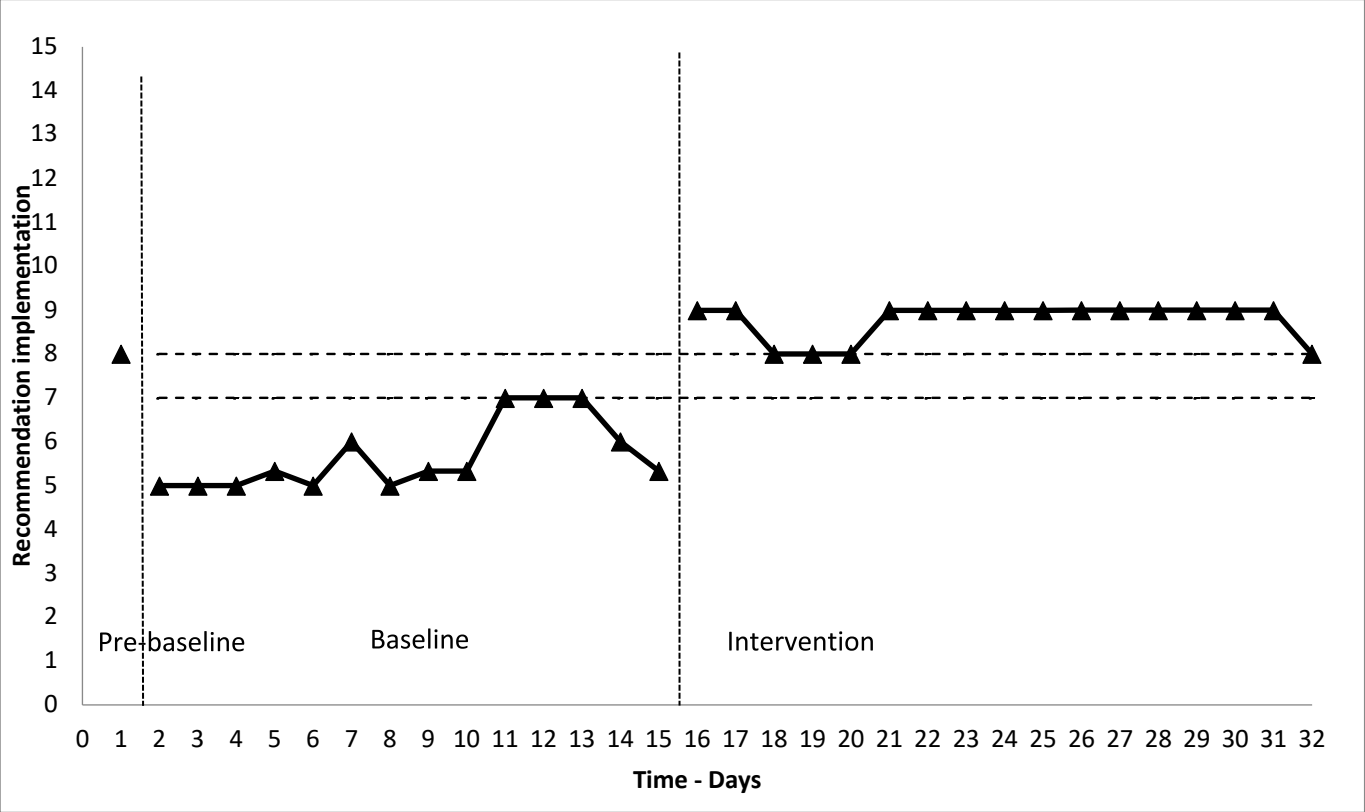
Participant 1: Overlap of data between phases for non-targeted recommendations



Participant 2: Overlap of data between phases for non-targeted recommendations
(no lines as all data overlap)



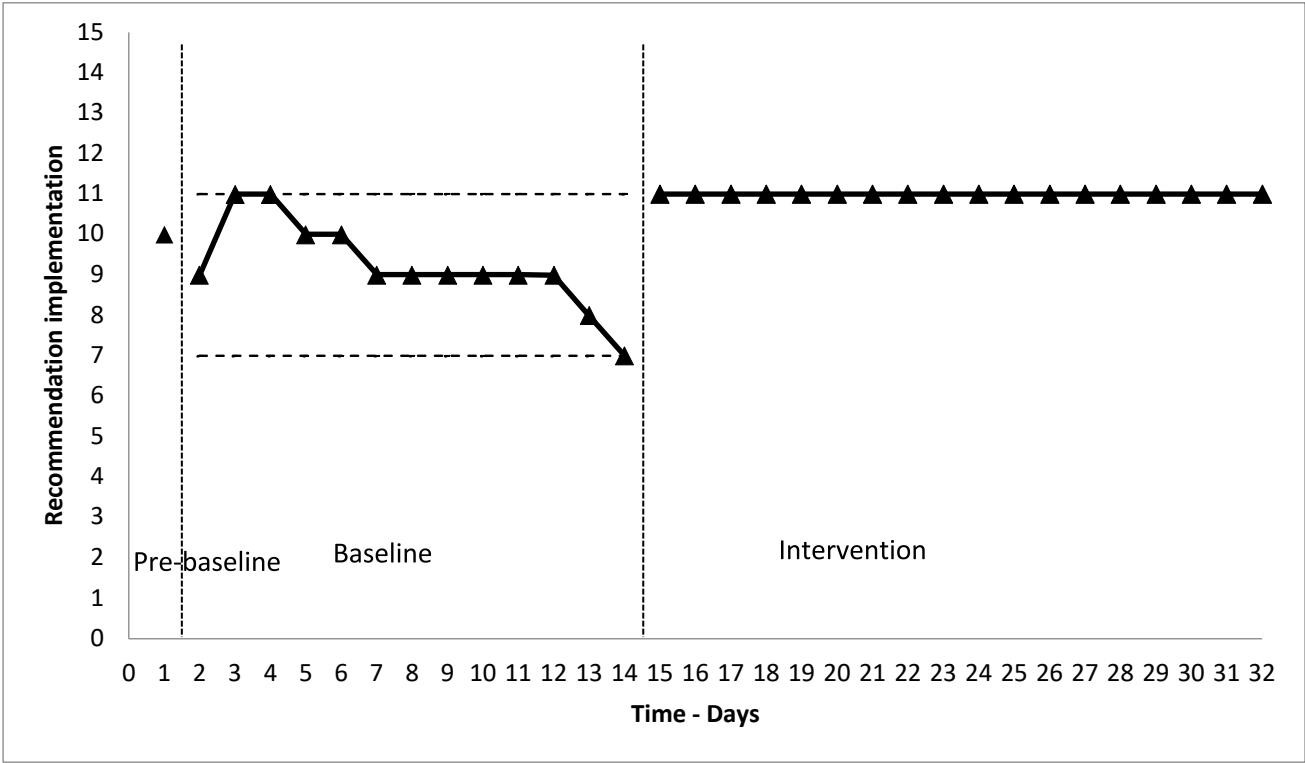
Participant 3: Overlap of data between phases for non-targeted recommendations



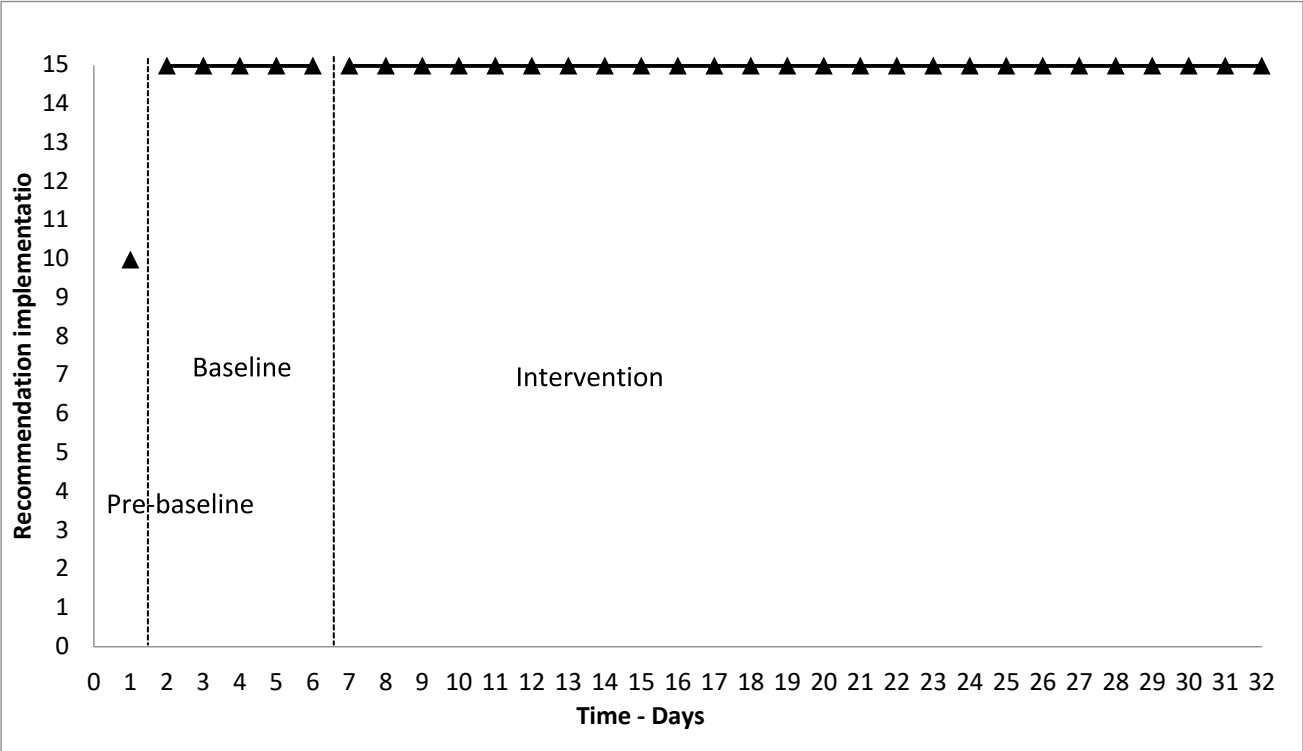
Participant 4: Overlap of data between phases for non-targeted recommendations

Figure 11

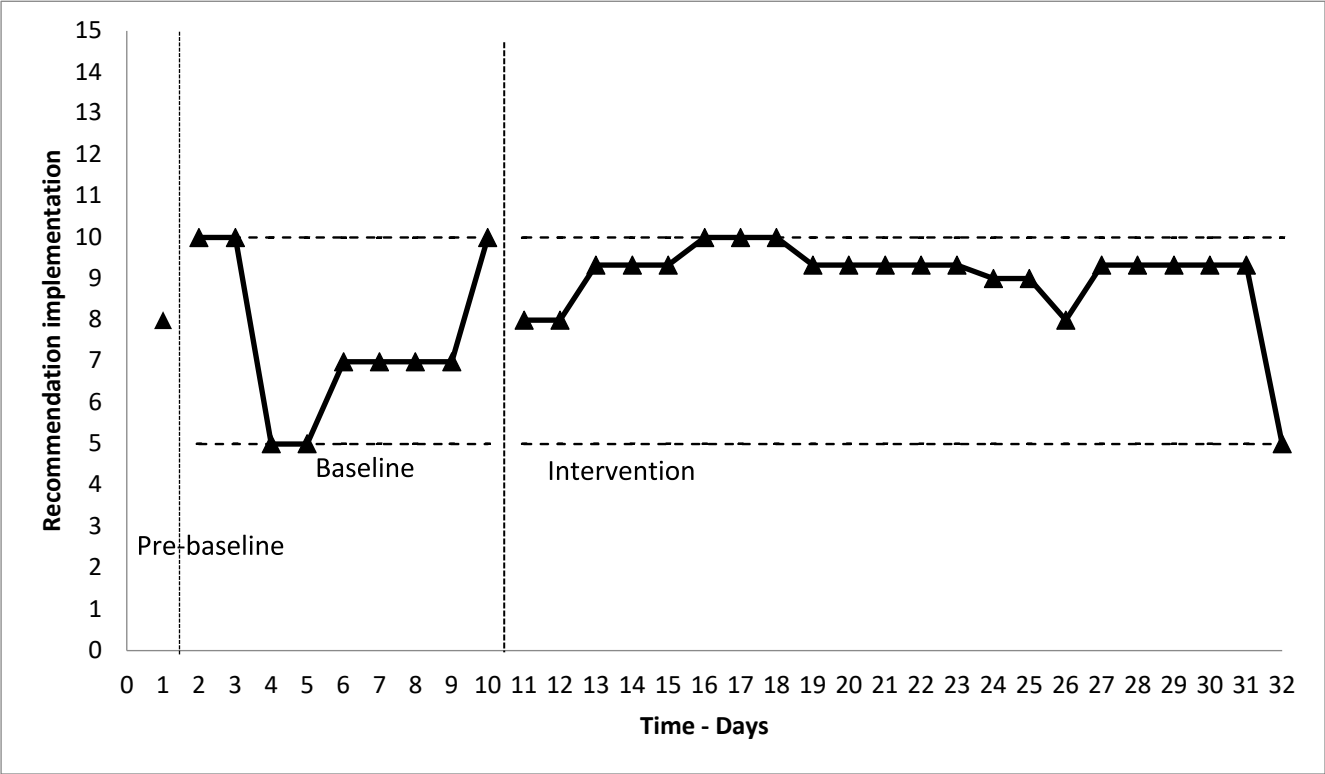
Implementation range across non-targeted recommendations for all participants



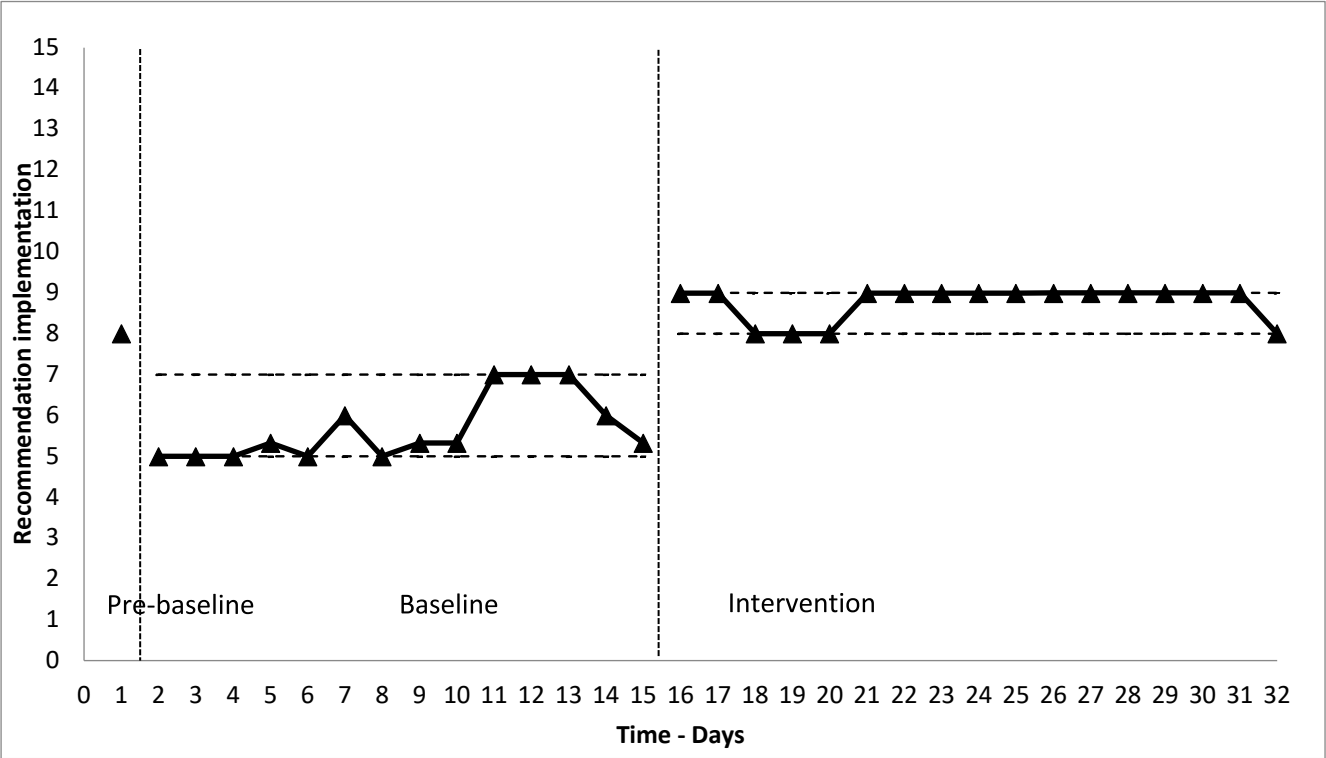
Participant 1: Implementation range across non-targeted recommendations



Participant 2: Implementation range across non-targeted recommendations



Participant 3: Implementation range across non-targeted recommendations



Participant 4: Implementation range across non-targeted recommendations

Appendix H: Questionnaire scores including subscales

Table 8

Subscale scores on the SDQ, BRIEF and Conners questionnaires

Questionnaire (Subscale)	Participant 1		Participant 2		Participant 3		Participant 4	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post
SDQ (raw score, interpretation)								
Emotional problems	7 (very high)	6 (high)	5 (high)	5 (high)	2 (close to average)	0 (close to average)	0 (close to average)	0 (close to average)
Conduct problems	4 (high)	3 (slightly raised)	3 (slightly raised)	2 (close to average)	4 (high)	2 (close to average)	5 (high)	4 (high)
Hyperactivity	10 (very high)	10 (very high)	2 (close to average)	6 (slightly raised)	10 (very high)	10 (very high)	10 (very high)	10 (very high)
Peer problems	3 (slightly raised)	4 (high)	1 (close to average)	2 (close to average)	3 (slightly lowered)	2 (close to average)	0 (close to average)	1 (close to average)

Prosocial	6 (low)	8 (close to average)	10 (close to average)	9 (close to average)	5 (very low)	8 (close to average)	5 (very low)	6 (low)
Total difficulties	25 (very high)	24 (very high)	12 (close to average)	16 (slightly raised)	20 (very high)	14 (slightly raised)	15 (slightly raised)	15 (slightly raised)
Impact	4 (very high)	4 (very high)	1 (slightly raised)	3 very high)	2 (high)	0 (close to average)	2 (high)	2 (high)
PedsQL (raw)								
Physical	14	15	3	4	2	1	0	0
Emotional	12	13	10	9	9	8	5	3
Social	8	10	2	3	8	4	1	3
School	9	5	12	10	10	9	9	6
General fatigue	16	12	12	11	3	0	3	4
Sleep fatigue	11	11	12	6	2	0	1	2
Cognitive fatigue	16	14	16	18	17	16	15	12

BRIEF (T-score)								
Inhibit	78	69	49	57	82	78	83	80
Shift	80	73	84	84	50	50	41	60
Emotional Control	76	76	75	68	62	65	63	65
BRI	82	76	71	71	69	68	67	72
Initiate	71	61	76	73	59	50	67	71
Working Memory	83	83	79	77	69	60	88	78
Planning/ Organising	70	67	78	66	56	54	90	80
Organising material	50	40	55	55	55	45	66	70
Monitoring	66	66	58	61	69	69	87	91

MI	71	67	73	69	64	57	87	83
GEC	77	72	74	71	67	62	81	81
<hr/>								
Conners (T-score)								
Inattention	75	68	81	68	76	69	>90	>90
Hyperactivity	84	75	57	66	86	>90	>90	>90
Learning problems	43	54	84	84	62	62	78	75
Executive Functioning	68	60	59	68	57	60	67	71
Defiance / aggression	59	79	52	52	84	45	54	54
Peer relations	77	89	45	51	54	62	45	45

Key: BRIEF – Behavior Rating Inventory of Executive Function; BRI – Behavioral Regulation Index; MI – Metacognition Index; GEC – Global Executive Composite

Appendix I: Dissemination and Author Instructions

Dissemination statement

This paper will be disseminated within The Clinical Neuropsychologist journal.

Instructions for Authors

Preparing Your Paper

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

Word Limits

Please include a word count for your paper. There are no word limits for papers in this journal.

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- There are no strict formatting requirements, but all manuscripts must contain the essential elements needed to evaluate a manuscript: abstract, author affiliation, figures, tables, funder information, and references. Further details may be requested upon acceptance.

- References can be in any style or format, so long as a consistent scholarly citation format is applied. Author name(s), journal or book title, article or chapter title, year of publication, volume and issue (where appropriate) and page numbers are essential. All bibliographic entries must contain a corresponding in-text citation. The addition of DOI (Digital Object Identifier) numbers is recommended but not essential.
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- Spelling can be US or UK English so long as usage is consistent.

Note that, regardless of the file format of the original submission, an editable version of the article must be supplied at the revision stage.

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In recent years, with the increasing recognition that there is a non-replication crisis in scientific publishing, many journals require that authors follow strict reporting guidelines to facilitate reproducibility of published studies. TCN reporting guidelines can be found on the link below. We encourage authors to print the TCN reporting guidelines checklist and use it to ascertain, in a point-by-point fashion, that all

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Formatting Checklist: What to Include

1. Author details. All authors of a manuscript should include their full name and affiliation on the cover page of the manuscript. Where available, please also include ORCiDs and social media handles (Facebook, Twitter or LinkedIn). One author will need to be identified as the corresponding author, with their email address normally displayed in the article PDF (depending on the journal) and the online article. Authors' affiliations are the affiliations where the research was conducted. If any of the named co-authors moves affiliation during the peer-review process, the new affiliation can be given as a footnote. Please note that no changes to affiliation can be made after your paper is accepted. Read more on authorship .
2. Abstract. Should contain a structured abstract of 250 words. A structured abstract should cover (in the following order): Objective: A brief statement of the purpose of the study. Method: A summary of the participants as well as descriptions of the study design, procedures, and specific key measures, to the extent that space allows. Results: A summary of the key findings. Conclusions: Clinical and theoretical implications of the findings. NOTE: If your manuscript is a critical review or a commentary, you can omit the Results portion of the abstract. However, retain that portion for systematic reviews and meta-analyses. Read tips on writing your abstract.

3. Graphical abstract (optional). This is an image to give readers a clear idea of the content of your article. It should be a maximum width of 525 pixels. If your image is narrower than 525 pixels, please place it on a white background 525 pixels wide to ensure the dimensions are maintained. Save the graphical abstract as a .jpg, .png, or .tiff. Please do not embed it in the manuscript file but save it as a separate file, labelled GraphicalAbstract1.

a. Video abstract (optional).

b. Find out how these can help your work reach a wider audience, and what to think about when filming.

4. Between 5 and 10 keywords. Read making your article more discoverable, including information on choosing a title and search engine optimization.

5. Funding details. Please supply all details required by your funding and grant-awarding bodies as follows:

For single agency grants

This work was supported by the [Funding Agency] under Grant [number xxxx].

For multiple agency grants

This work was supported by the [Funding Agency #1] under Grant [number xxxx]; [Funding Agency #2] under Grant [number xxxx]; and [Funding Agency #3] under Grant [number xxxx].

6. Disclosure statement. This is to acknowledge any financial interest or benefit that has arisen from the direct applications of your research. Further guidance on what is a conflict of interest and how to disclose it .

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8. Data deposition. If you choose to share or make the data underlying the study open, please deposit your data in a recognized data repository prior to or at the time of submission. You will be asked to provide the DOI, pre-reserved DOI, or other persistent identifier for the data set.
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11. Figures. Figures should be high quality (1200 dpi for line art, 600 dpi for grayscale and 300 dpi for colour, at the correct size). Figures should be supplied in one of our preferred file formats: EPS, PS, JPEG, TIFF, or Microsoft Word (DOC or DOCX) files are acceptable for figures that have been drawn in Word. For information relating to other file types, please consult our Submission of electronic artwork document.
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